CLINICAL NOTE

Persistent Foramen of Huschke Mimicking a Branchial Cleft Anomaly

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A persistent foramen of Huschke, or foramen tympanicum, results from the defective ossification of the anteroinferior aspect of the tympanic portion of the temporal bone. We describe 2 girls, aged 5 and 6 years, with histories of recurrent cervical abscesses, draining submandibular sinuses, and defective tympanic plates adjacent to the tympanic ring. Surgical resection of the fistulous tract extending from the submandibular triangle to the bony ear canal successfully controlled the symptoms.

REPORT OF CASES

CASE 1

A 5-year-old girl presented to a tertiary care pediatric otolaryngology outpatient clinic with a history of swelling in the anterior aspect of the left side of her neck. She had the sudden onset of left submandibular swelling and erythema 5 weeks before presentation, which then developed into cutaneous drainage and improved after the administration of intravenous antibiotics at a different institution. Her medical history was remarkable for a tympanoplasty and mastoidectomy that had been performed on that same side 1 year earlier, with a diagnosis of chronic suppurative otitis media. On physical examination, she had a draining sinus that was surrounded by granulation tissue right above the hyoid but was afebrile and nontoxic. Computed tomography (CT) revealed a tract extending from the cervical skin up into the bony external auditory canal (EAC) through a dysmorphic anteroinferior wall (**Figure 1**). The patient was then taken to the operating suite, and the tract was resected through a superficial parotidectomy approach, with facial nerve identification and preservation. The tract extended into the styloid process, which was amputated and packed with a fat plug. Examination of the surgical specimen revealed a 3-cm-long fibrotic tract with acute and chronic inflammation. There were no complications or evidence of recurrence during the 1-year follow-up period.

CASE 2

A 6-year-old girl presented to the emergency department with the sudden onset of right submandibular swelling, tenderness, and erythema. Associated symptoms also included right-sided otalgia and otorrhea. Her medical history was notable for 2 previous procedures for excision of right submandibular cysts. On physical examination, she had a draining sinus approximately 1 cm below the right angle of the mandible as well as substantial otorrhea and granulation tissue occluding the ear canal. A CT of the neck with contrast showed significant inflammatory changes in the parotid gland, temporomandibular joint, and parapharyngeal space. A superficial parotid fluid collection was drained, and oral and ototopical antibiotic therapy was initiated. After the infection subsided, the results of magnetic resonance imaging confirmed the CT findings of a fistulous tract extending from the submandibular area into the medial aspect of the EAC through a dysmorphic styloid bone and an anteroinferior wall (**Figure 2**). The patient was also

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taken to the operating suite, and the tract was resected through a similar superficial parotidectomy approach, with facial nerve identification and preservation. The styloid process was also amputated, and the lumen at its insertion was circumferentially cauterized and packed with a sterile absorbable gelatin sponge (Gelfoam; Pharmacia & Upjohn Co) and an absorbable hemostat (Surgicel; Ethicon 360). There was no evidence of recurrence during the 18-month follow-up period. Histopathologic examination of the surgical specimen revealed a 3.5-cm-long fibroadipose tract with acute and chronic inflammation.

COMMENT

Work[1] has described 2 categories of first branchial cleft anomalies based on histologic features and proposed embryology. Type 1 lesions are usually located around the pinna, run parallel to the EAC, and are ectodermal in origin, while type 2 lesions possess 2 germ cell layer derivatives (ectoderm and mesoderm), mostly skin and cartilage. Type 2 lesions are often located in the parotid or submandibular areas, above the level of hyoid, and extend up into the EAC in a variable course relative to the main trunk and branches of the facial nerve. Several variations of type 2 anomalies have been described, including communication with the tympanic cavity and nasopharynx; however, to our knowledge, there is no description of a fistulous tract from the skin going into the EAC directly through a cavity within the styloid process.[2] The styloid bone is widely regarded as a derivative of Reichert cartilage, which is a structure from the second branchial arch that also gives rise to the stapes, the styloid ligament, and the lesser cornu and superior part of the hyoid bone and is therefore not involved in first branchial arch anomalies. An unusual cavity of the styloid process has been described by Prescher et al,[3] who treated an adult patient with a history of chronic otorrhea, multiple otologic procedures, recurring inflammation, and abscesses of the retromandibular and submandibular regions. In their case, only the cranial aspect of the styloid process was significantly dilated and in continuity with the tympanic cavity.

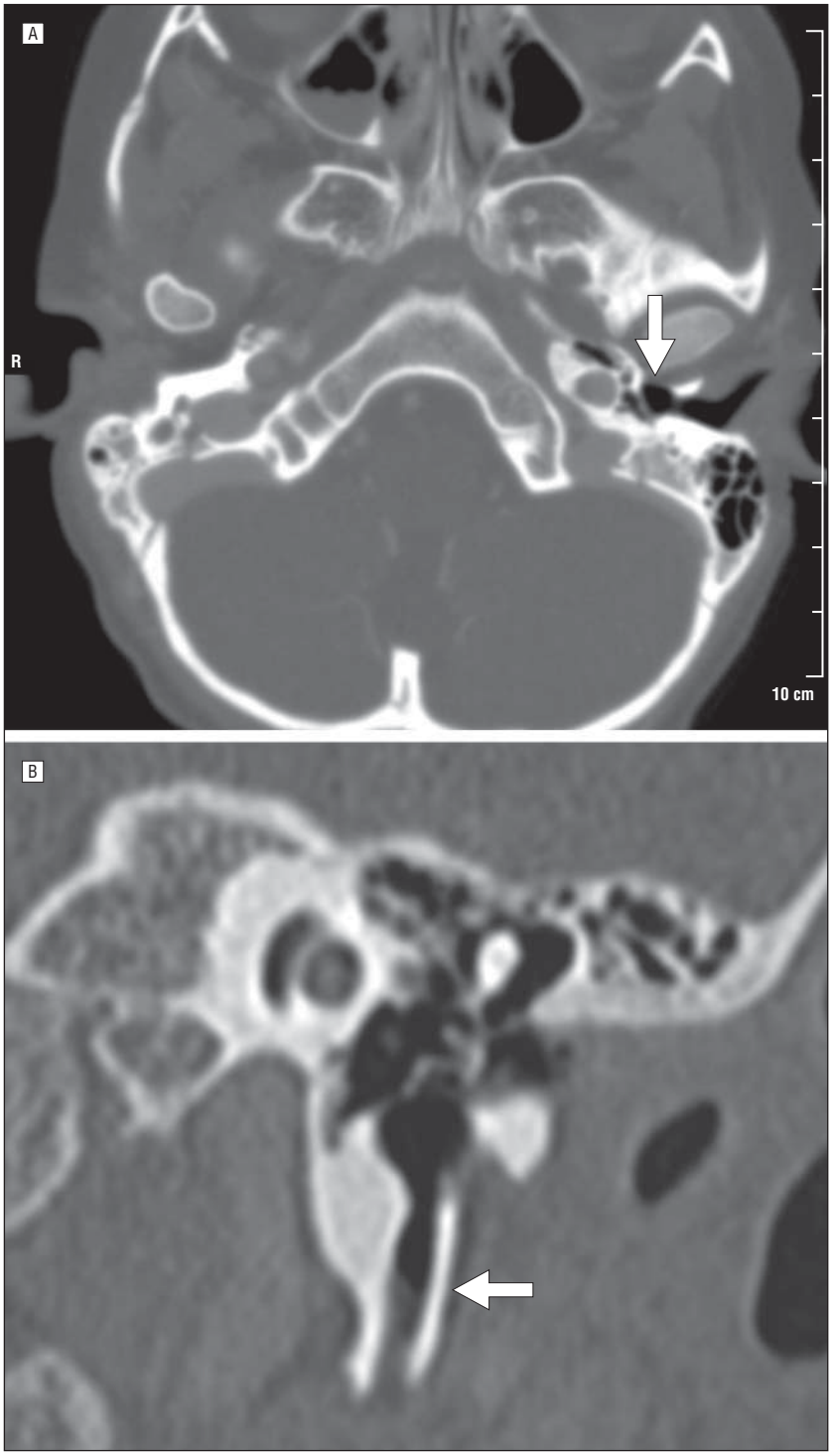

Figure 1. Case 1. A postcontrast computed tomogram depicting an anteroinferior external auditory canal defect (A, arrow) and a coronal cut showing a dysmorphic styloid process (B, arrow).

The location and extent of the bony defects in our 2 patients, at the anteroinferior aspect of the medial portion of the EAC, are consistent with a persistent foramen of Huschke (or foramen tympanicum), an anatomical variation in the tympanic portion of the temporal bone, which is also listed in the differential diagnosis of recurrent infections of the ear canal and upper neck area. Yetiser et al[4] reported a

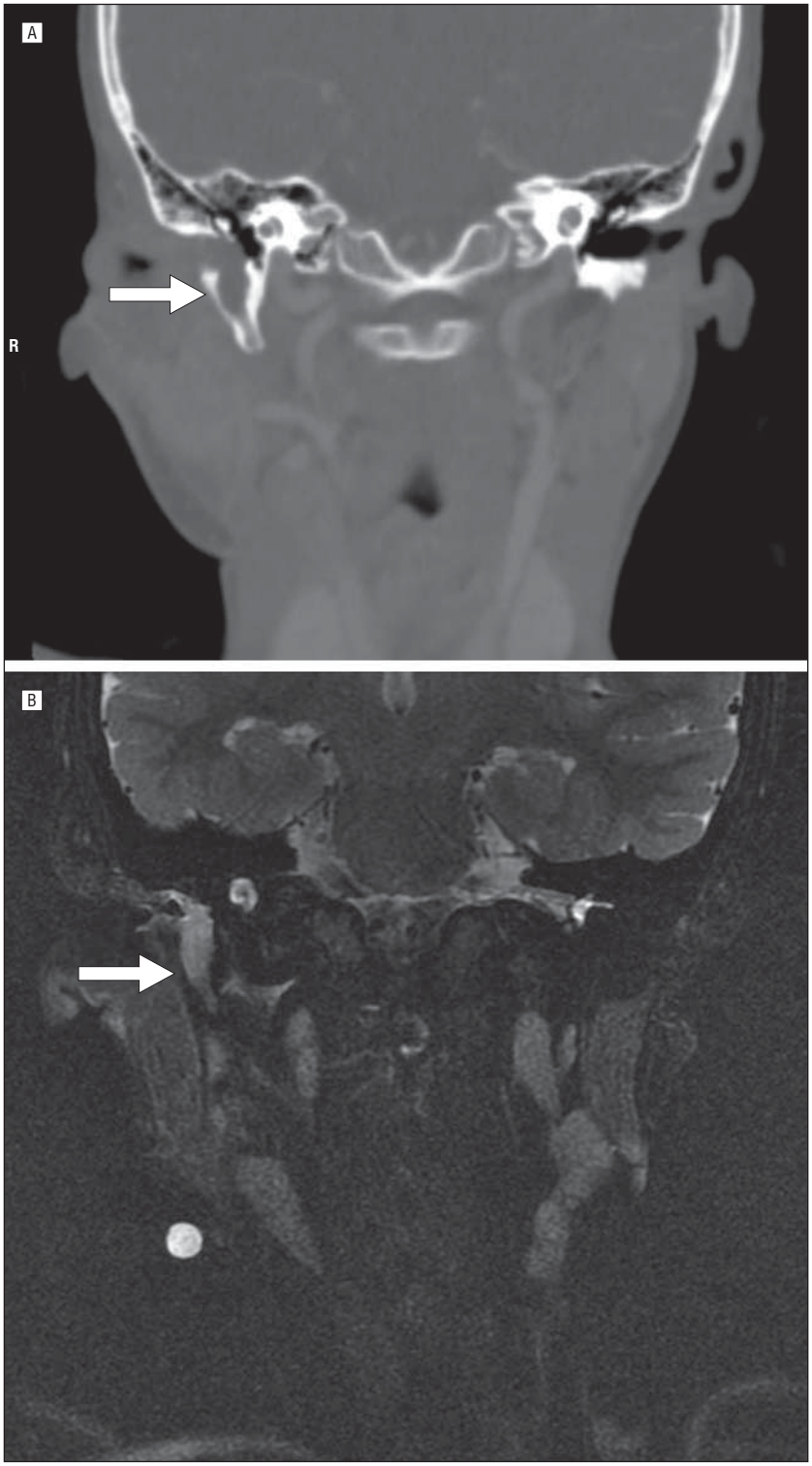

Figure 2. Case 2. A postcontrast coronal computed tomogram showing a dysmorphic styloid process and bony ear canal defect (A, arrow) and a T2-weighted fat-suppressed magnetic resonance image showing a fistulous tract (B, arrow).

similar defect in association with recurrent otorrhea and a parotid gland mass in an adult patient. The tympanic bone, which contributes to the formation of the ear canal and tympanic cavity, develops from a membranous ossification process around the tympanic membrane and fuses with the squamous portion of the temporal bone. The tympanic bone is still incompletely developed at birth, and a foramen may persist in the anteroinferior aspect of the EAC.[5,6] The persistence of such foramina is estimated to occur in approximately 7% of individuals and may predispose to temporomandibular abnormalities or fistulas between the ear canal and the parotid gland.[7] Salivary otorrhea during mastication and resultant otologic complications have been previously reported.[8,9] In our 2 patients, this dysmorphic feature served as a site of chronic infection that tracked caudally to the neck, resulted in recurrent abscesses of the parotid and submandibular areas, and later caused fistulization that mimicked a type 2 first branchial cleft anomaly. The management of the lesion is dictated by the symptoms, and in our cases, we deemed it necessary to excise the fistulous tract and diseased salivary tissue through a cervical approach given the presence of a skin fistula and the history of recurring neck abscesses that had been previously incised and drained. Pure closure of the ear canal defect may be appropriate in selected cases.

In conclusion, to our knowledge, this article gives the first description of a symptomatic foramen of Huschke in children, and we believe that such lesions should be among the differential diagnoses of congenital anomalies involving the ear canal in the pediatric population.

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