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Case report

Turicella otitidis hypoglossal nerve stimulator device associated infection

Angela McGaugh [a,*], William Russell [b], Erin Boswell [c]

[a] Tulane University School of Medicine Department of Internal Medicine, New Orleans, LA, USA
[b] Tulane University School of Medicine Department of Medicine, Section of Infectious Diseases, New Orleans, LA, USA
[c] Southeast Louisiana Veterans Health Care System, Section of Infectious Diseases, New Orleans, LA, USA

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ABSTRACT

71-year-old male with history of obstructive sleep apnea presented with persistent drainage from the surgical incision site over the recently implanted hypoglossal nerve stimulator. Wound cultures from device pocket identified the pathogen as *Turicella otitidis*. Clinical course included treatment with broad-spectrum intravenous antibiotics and device explantation. This case is the first known *T. otitidis* device associated infection.

Background

Turicella otitidis or *Corynebacterium otitidis* is an aerobic, gram-positive bacillus that is a part of the normal ear microbiome but has also been implicated as an otopathogen [1,2]. Its clinical correlation to human disease is primarily through cases of otitis media and externa in pediatric populations; however, there are some reports that indicate it could have expanded pathogenic potential outside of the auditory canal [3–6]. There have been reports implicating *T. otitidis* in palmoplantar eczema, endophthalmitis, microbial keratitis, and even bacteremia in an immunocompromised patient [3–6]. This pathogen has been becoming more easily identifiable with advances in rapid molecular testing with techniques such as matrix-assisted laser desorption/ionization time of flight mass spectrometry (MALDI-TOF) [7]. To our knowledge, this case is the first reported case of an *T. otitidis* device associated infection.

Case presentation

A 71-year-old male with past medical history significant for obstructive sleep apnea underwent implantation of an Inspire® hypoglossal nerve stimulator. The post-operative course was uncomplicated until postop day #14 when the patient noted increased serous drainage from his chest surgical incision. The patient was promptly seen in Otorhinolaryngology outpatient clinic, where a superficial abscess was found and ~ 10 ccs of serosanguinous fluid were expressed from the device pocket. Patient was prescribed a 7-day course of Trimethoprim/Sulfamethoxazole (TMP/SMX); however, purulent discharge from the chest wall surgical site persisted on postop day #17. Drainage from the device was cultured after 3 days of TMP/SMX use. This wound culture grew gram-variable rods. VITEK ® mass spectrometry identified this organism as *T. otitidis*. Patient then presented to the emergency department on postop day #21 with complaints of drainage from not only the device pocket, but also from his neck incision. On exam, patient was afebrile and hemodynamically stable with no leukocytosis. On exam, there was noted to be induration with purulent drainage from both the device pocket and right lateral neck incisions. Both were packed with gauze, and blood and wound cultures were sent from both neck and chest wall surgical sites. Patient was admitted and started on empiric IV antibiotic therapy with vancomycin and piperacillin-tazobactam. Neck surgical site and device pocket cultures on admission also grew *T. otitidis*. Blood cultures drawn on hospital admission were negative. Computed Tomography Scan of neck soft tissue and thorax with contrast was notable for soft tissue thickening and stranding around implanted chest device as well as associated pockets of subcutaneous gas and enlarged cervical lymph nodes (Fig. 1). On postop day #22, patient underwent surgical removal of the hypoglossal implant plus incision and drainage of chest and neck surgical sites with Jackson-Pratt drain placement. Intraoperatively, wound cultures were taken from both neck and chest wall surgical sites. Intraoperative cultures taken from neck and chest wall sites grew normal skin flora with no isolation of anaerobes. Patient was deescalated to vancomycin alone once blood cultures and operative wound cultures showed no gram-negative growth. Patient was then discharged three days after device explantation with six days of amoxicillin/clavulanic acid for total 10-

* Corresponding author.
E-mail address: amcgaugh@tulane.edu (A. McGaugh).

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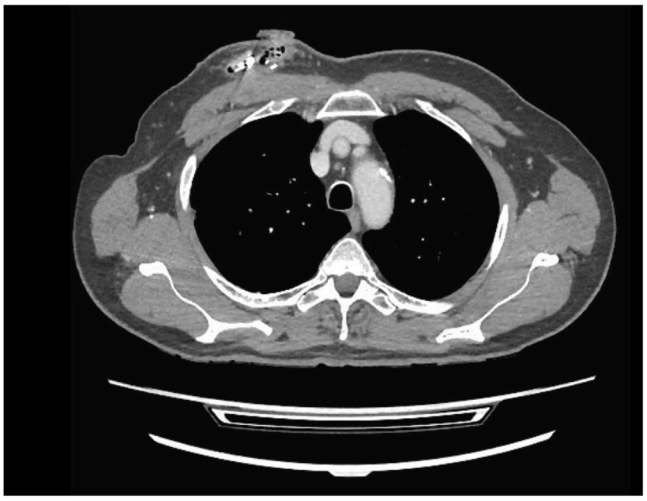

Fig. 1. Computed tomography thorax without contrast showing inflammation around chest device prior to explantation.

day antibiotic course. Patient was evaluated in outpatient Infectious Disease clinic one week after hospital discharge. Patient had completed oral antibiotic course. Patient reported persistent clear drainage from chest wall surgical wound, which had been present but decreased since hospital discharge. Patient denied any systemic signs of infection or increased pain, purulent drainage, swelling, or erythema around surgical sites. Patient was instructed to continue to monitor surgical incision sites and discharged from clinic with no further antibiotics. On telephone follow up, patient had denied any recurrence of drainage three months after discharge.

Discussion

T. otitidis is an atypical organism outside of the auditory tract and has only been implicated in one case report as a post-surgical infection outside of the auditory tract [4]. This is the first known device associated infection with this pathogen. A 10-year FDA review of adverse events with Inspire hypoglossal nerve stimulator showed that the most common adverse event was infection, with majority of cases leading to explantation [8]. Presence of *T. otitidis* in multiple purulent cultures spanning different locations and points in time is highly suggestive of *T. otitidis* being the pathogen responsible for this infection. *T. otitidis* has previously been described with high rates of TMP/SMX resistance, which is consistent with the patient's prior failure of antibiotic therapy [9]. This case may not have been identified if the patient was treated with an alternative initial therapy and suggests the need to consider otolaryngeal flora as potential etiologies. Many *T. otitidis* infections were likely underdiagnosed or missed prior to 16S rRNA sequencing as it was phenotypically difficult to distinguish it from other coryneforms in culture [9–11]. The expansion of MALDI-TOF systems continues to provide more concise identification of an ever-expanding list of pathogens. More investigation into the pathogenicity of *T. otitidis* may be indicated and assessed going forward.

CRediT authorship contribution statement

Erin Boswell: Writing – original draft, Writing – review & editing. **William Russell:** Writing – original draft, Writing – review & editing. **Angela McGaugh:** Writing – original draft, Writing – review & editing.

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Ethical approval

Written informed consent was obtained from the patient for publication of this case report and accompanying image.

Consent

A copy of the written consent is available for review by the Editor-in-Chief of this journal on request.

Declaration of Competing Interest

None. There are no declaration of interest among all authors involved.

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Author contributions

All authors were involved in the writing, revision, and submission of this case report. Dr. Angela McGaugh was the primary author involved in writing the majority of case report. Dr. William Russell was supervising infectious disease fellow on case report who assisted in writing and editing. Dr. Erin Boswell was the overseeing infectious disease attending and involved in revising and editing drafts.

References

[1] Stroman DW, Roland PS, Dohar J, Burt W. Microbiology of normal external auditory canal. Laryngoscope 2001;111:2054–9. https://doi.org/10.1097/00005537-200111000-00035.
[2] Funke G, Stubbs S, Altwegg M, Carlotti A, Collins MD. *Turicella otitidis* gen. nov., sp. nov., a coryneform bacterium isolated from patients with otitis media. Int J Syst Bacteriol 1994;44:270–3. https://doi.org/10.1099/00207713-44-2-270.
[3] Koumaki D, Koumaki V, Boumpoucheropoulos S, Katoulis A, Bitados P, Stefanidou M, et al. *Turicella otitidis* as an unusual agent causing palmoplantar eczema: an emerging pathogen. Eur J Case Rep Intern Med 2020;7(2):001458. https://doi.org/10.12890/2020_001458 [PMID: 32133318; PMCID: PMC7050971].
[4] Mammo DA, Watson D, Armbrust KR. Post-intravitreal injection endophthalmitis secondary to *Turicella otitidis*: a case report. BMC Ophthalmol 2020;20(1):142. https://doi.org/10.1186/s12886-020-01412-1 [PMID: 32276621; PMCID: PMC7146956].
[5] Priyadarshini SR, Behera HS, Sahu S, et al. *Turicella otitidis*: a rare agent causing microbial keratitis. BMJ Case Rep CP 2021;14:e241371..
[6] Li D, Shan W, Dai C, Kan J, Li M, Yin M. Bacteremia caused by *Turicella otitidis* in a patient with diffuse large B-cell lymphoma. Clin Lab 2020;66(3). https://doi.org/10.7754/Clin.Lab.2019.190641 [PMID: 32162876].
[7] Singhal N, Kumar M, Kanaujia PK, Virdi JS. MALDI-TOF mass spectrometry: an emerging technology for microbial identification and diagnosis. Front Microbiol 2015;6:791. https://doi.org/10.3389/fmicb.2015.00791 [PMID: 26300860; PMCID: PMC4525378].
[8] Bestourous DE, Pasick LJ, Benito DA, Zapanta PE. Adverse events associated with the Inspire implantable hypoglossal nerve stimulator: a MAUDE database review. Am J Otolaryngol 2020;41(6):102616. https://doi.org/10.1016/j.amjoto.2020.102616 [Epub 2020 Jun 25. PMID: 32645535].
[9] Lappan R, Jamieson SE, Peacock CS. Reviewing the pathogenic potential of the otitis-associated bacteria *Alloiococcus otitidis* and *Turicella otitidis*. Front Cell Infect Microbiol 2020;10:51. https://doi.org/10.3389/fcimb.2020.00051 [PMID: 32117817; PMCID: PMC7033548].
[10] Funke G, Stubbs S, Altwegg M, Carlotti A, Collins MD. *Turicella otitidis* gen. nov., sp. nov., a coryneform bacterium isolated from patients with otitis media. Int J Syst Bacteriol 1994;44:270–3. https://doi.org/10.1099/00207713-44-2-270.
[11] Von Graevenitz A, Funke G. *Turicella otitidis* and *Corynebacterium auris*: 20 years on. Infection 2014;42:1–4. https://doi.org/10.1007/s15010-013-0488-x.