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Osteomyelitis due to Stenotrophomonas maltophilia treated with trimethoprim-sulfamethoxazole monotherapy

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Keywords:	Stenotrophomonas maltophilia is an opportunistic infection commonly encountered in various systems of the human body and has been noted to be a rare cause of osteomyelitis. This report examines a rare case of osteomyelitis of the foot caused by S. maltophilia from a poorly managed foot wound and highlights the successful treatment of this condition with trimethoprim-sulfamethoxazole monotherapy.
Osteomyelitis Treatment Sternotrophomonas maltophilia Trimethoprim-sulfamethoxazole	

Introduction

Stenotrophomonas maltophilia is an opportunistic aerobic gramnegative bacillus that is commonly recognized as a respiratory pathogen, but the organism has also been associated with other infections including urinary tract infections, respiratory tract infections, gastrointestinal, biliary system, nervous system and spinal cord, skin, soft tissue and bone infections and implant infections [1–3]. Only very few cases of osteomyelitis caused by Stenotrophomonas maltophilia have been reported [7]. Although, literature reports that incidence of Stenotrophomonas maltophilia infections are on the rise [3,4]. The index case describes a patient with a poorly managed foot wound who developed osteomyelitis due to S. maltophilia with successful treatment.

Case report

52-year-old male with medical history of hypertriglyceridemia presenting to the emergency department with worsening erythema, pain and swelling of the right great toe. The patient sustained a wound after removing the skin from the plantar side of his right big toe that resulted in a wound about a year prior to presentation. Patient continued selftreatment with intermittent soaking with hydrogen peroxide and Neosporin application. No prior use of systemic antibiotics was reported. The patient did not have any acute medical follow-up despite noticing a tracking hole underneath his right great toe. Patient noticed progressively worsening swelling, erythema of the right great toe with minimal drainage prompting presentation to the emergency department. Patient denied having any fever, chills, or rigor. On presentation to the emergency department, temperature was 98.4°F, pulse 105/min, respiratory rate of 18/min, pulse oximetry 99% on room air. Initial x-ray of the foot showed no radiographic evidence of bony destruction. MRI of the right foot showed soft tissue ulceration at the level of the interphalangeal joint of the first digit with septic arthritis and osteomyelitis of the phalanges at the interphalangeal joint, infectious tenosynovitis about the flexor hallucis tendon tracking proximally to the level of the sustentaculum tali. Patchy marrow edema and enhancement within the plantar aspect of the sesamoids which are in proximity to the flexor hallucis tendon concerning for early osteomyelitis (Figs. 1, 2 and 3). Laboratory findings include an erythrocyte sedimentation rate of 77 mm/h (normal range, 0–30 mm/h), C-reactive protein concentration was 91.7 mg/dL (normal range, 0–1.0 mg/dL).

On examination, right foot plantar hallux ulcer post debridement measured $1 \text{ cm} \times 1.6 \text{ cm} \times 0.6 \text{ cm}$ with positive probe to bone and proximal tracking with mild purulence expressed, with significant proximally tracking edema and erythema.

Tissue culture from debridement grew Stenotrophomonas maltophilia identified by matrix-assisted laser desorption/ionization time-of-flight (MALDI-ToF) mass-spectrometry. The isolate was susceptible to levofloxacin (MIC, $\leq 0.5 \ \mu$ g/mL), trimethoprim-sulfamethoxazole (MIC, $\leq 0.5/9.5 \ \mu$ g/mL) by micro scan and minocycline by Kirby-Bauer testing.

A regimen of vancomycin and ceftriaxone was started empirically, later switched to levofloxacin. The patient was eventually discharged on sulfamethoxazole-trimethoprim to complete a course of 6 weeks with weekly wound care follow-up. Patient showed progressive improvement and healing in right hallux wound.

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Case report





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Fig. 1. (showing patchy decreased T1 marrow).



Fig. 2. (Showing marrow edema on the STIR sequence).



Fig. 3. (Showing enhancements).

Discussion

Risk factors like immunosuppression, increased placement of catheters and other devices, increased use of antibiotics have been implicated in infections with S. maltophilia [2, 4, 5]. The index patient did not have a known history of immunosuppression or other comorbidities to suggest immunosuppression, he did however continue home treatment of his toe wound leading to infection over a prolonged period. S. maltophilia is known to exhibit intrinsic resistance to a wide range of antibiotics through expression of beta-lactamases, zinc containing penicillinase, cephalosporinase, and efflux pumps for quinolones [1-3]. Culture reports show the organism isolated from the index patient was susceptible to trimethoprim-sulfamethoxazole, levofloxacin, and minocycline. Although the patient was initially started on levofloxacin, he was subsequently switched to trimethoprim-sulfamethoxazole, as the preferred drug of choice in keeping with literature [2,3]. Strong studies to compare effectiveness of microbial agents and treatment of S. maltophilia are lacking, and majority of evidence are from case reports [10]. Like the current case, a study that reviewed antimicrobial susceptibilities for S. maltophilia also found trimethoprim-sulfamethoxazole, levofloxacin, and minocycline as the most effective antimicrobial agents [8]. Another study also described trimethoprim-sulfamethoxazole, doxycycline and tigecycline as effective single agents, and trimethoprim-sulfamethoxazole plus ceftazidime and trimethoprim-sulfamethoxazole plus ticarcillin/clavulanate as effective synergistic agents [9]. Some recommendations suggest using trimethoprim-sulfamethoxazole synergistically ticarcillin-clavulanate [6,7]. Manufacturing of ticarcillin-clavulanate has been discontinued in the United States and some issues with confidence in Minimal inhibitory concentration (MIC) interpretation reproducibility of ceftazidime and levofloxacin have also been noted [10]. The infectious disease Society of America suggests using trimethoprim-sulfamethoxazole, minocycline, tigecycline, levofloxacin or cefiderocol monotherapy for mild infection, combination of trimethoprim-sulfamethoxazole and combination minocvcline therapy, initiation of trimethoprim-sulfamethoxazole with addition of a second agent if there is delay in clinical improvement for moderate to severe infections [10]. TMP-SMX was the drug of choice in the index case because literature has shown that despite the in vitro advantage of being bacteriocidal and having a lipophilic structure, flouroquinolones have been known to develop resistance during therapy [6]. Other studies have also shown more frequent development of resistance to therapy with levofloxacin in comparison to TMP-SMX, which is attributed to lower barrier to reflux pumps or mutation of drug target mediated resistance [11,12]. TMP-SMX is also suggested to be the only antimicrobial to which development of resistance during treatment has not been shown [6]. successfully treated This patient was with trimethoprim-sulfamethoxazole monotherapy and weekly wound care. In conclusion, this case shows that osteomyelitis due to Stenotrophomonas maltophilia can be successfully treated with trimethoprim-

trophomonas maltophilia can be successfully treated with trimethoprimsulfamethoxazole monotherapy, decision on what antimicrobial to use should be determined by susceptibility testing. More robust studies are needed to improve evidence and possibly create guidelines for treatment of this organism.

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Consent

Informed consent was obtained from the patient for publication of this case report

Author statement

This work was solely done and submitted by Nonso Osakwe, MD, MPH.

CRediT authorship contribution statement

This work was solely done by Nonso Osakwe, MD. Informed consent was obtained from the patient for publication of this case report.

Declaration of Competing Interest

The author has no conflict of interest to declare.

References

- Brooke JS. Advances in the microbiology of stenotrophomonas maltophilia. Clin Microbiol Rev 2021;34(3):e0003019. https://doi.org/10.1128/CMR.00030-19. Epub 2021 May 26. PMID: 34043457; PMCID: PMC8262804.
- [2] Umar Z, Ilyas U, Ashfaq S, Bhangal R, Nassar M. Stenotophomonas maltophilia as not just a mere colonozer: two cases of urinary tract infection and multidrugresistant respiratory infection. Cureus 2022;14(3):e23541. https://doi.org/ 10.7759/cureus.23541. PMID: 35494901; PMCDD: PMC9045462.
- [3] Falagas ME, Valkimadi PE, Huang YT, Matthaiou DK, Hsueh PR. Therapeutic options for Stenotrophomonas maltophilia infections beyond co-trimoxazole: a systematic review. J Antimicrob Chemother 2008;62(5):889–94. https://doi.org/ 10.1093/jac/dkn301. Epub 2008 Jul 28. PMID: 18662945.
- [4] Ahmed M, Syed R, More YI, Basha SI. Stenotrophomonas skull base osteomyelitis presenting as necrotizing otitis externa: Unmasking by CT and MRI-case report and review. Radio Case Rep 2019;14(10):1241–5. https://doi.org/10.1016/j. radcr.2019.07.018. PMID: 31462947; PMCID: PMC6706613.

- [5] Adsul NM, Panigrahi V, Acharya S, Kalra KL, Chahal RS. Stenotrophomonas maltophilia spondylodiscitis following lumbar microdiscectomy mimicking a cotton granuloma: A case report and literature review. Surg Neurol Int 2020;11:28. https://doi.org/10.25259/SNI_23_2020. PMID: 32123616; PMCID: PMC7049875.
- [6] Landrum ML, Conger NG, Forgione MA. Trimethoprim-sulfamethoxazole in the treatment of Stenotrophomonas maltophilia osteomyelitis. Clin Infect Dis 2005;40 (10):1551–2. https://doi.org/10.1086/429730.
- [7] Chesnutis Edward J, Ng Alan, Kruse Dustin, Stone Paul A. Stenotrophomonas maltophilia: a rare case of osteomyelitis after an open distal tibial fracture. ISSN 1067-2516 J Foot Ankle Surg 2018;Volume 57(Issue 5):1037–41. https://doi.org/ 10.1053/j.jfas.2018.03.001.
- [8] Chung HS, Hong SG, Kim YR, Shin KS, Whang DH, Ahn JY, Park YJ, Uh Y, Chang CL, Shin JH, Lee HS, Lee K, Chong Y. Antimicrobial susceptibility of stenotrophomonas maltophilia isolates from Korea, and the activity of antimicrobial combinations against the isolates. J Korean Med Sci 2013;28(1): 62–6. https://doi.org/10.3346/jkms.2013.28.1.62. Epub 2013 Jan 8. PMID: 23341713; PMCID: PMC3546106.
- [9] Gülmez D, Cakar A, Sener B, Karakaya J, Hasçelik G. Comparison of different antimicrobial susceptibility testing methods for Stenotrophomonas maltophilia and results of synergy testing. J Infect Chemother 2010;16(5):322–8. https://doi.org/ 10.1007/s10156-010-0068-2. Epub 2010 May 7. PMID: 20449623.
- [10] Tamma P.D., Aitken S.L., Bonomo R.A., Mathers A.J., van Duin D., Clancy C.J. Infectious Diseases Society of America Guidance on the Treatment of AmpC β-lactamase-Producing Enterobacterales, Carbapenem-Resistant Acinetobacter baumannii, and Stenotrophomonas maltophilia Infections. Infectious Diseases Society of America 2022; Version 2.0. Available at https://www.idsociety.org/ practice-guideline/amr-guidance-2.0/.
- [11] Garrison MW, Anderson DE, Campbell DM, et al. Stenotrophomonas maltophilia: emergence of multidrug-resistant strains during therapy and in an in vitro pharmacodynamic chamber model. Antimicrob Agents Chemother 1996;40(12): 2859–64. https://doi.org/10.1128/AAC.40.12.2859.
- [12] Gibb J, Wong DW. Antimicrobial treatment strategies for stenotrophomonas maltophilia: a focus on novel therapies. Published 2021 Oct 9 Antibiotics 2021;10 (10):1226. https://doi.org/10.3390/antibiotics10101226.