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Neisseria elongata osteomyelitis: Literature review and case report in a 63-year-old male presenting with progressive right-handed redness, swelling and pain

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ABSTRACT

INTRODUCTION: *Neisseria elongata*, a gram-negative rod, has been identified in 23 cases of pathogenesis in the literature, of which only 2 were osteomyelitis. Here we report the third known case of isolated *N. elongata* cultures in a digital osteomyelitis.

PRESENTATION OF CASE: A 63-year-old male with peripheral vascular disease and lower extremity wounds presented to his vascular surgeon with right-handed redness and pain requiring urgent surgical intervention. Irrigation, debridement, and partial thumb amputation were performed, followed by treatment with a 6-week course of antibiotics.

DISCUSSION: *N. elongata* is a rare cause of osteomyelitis and unfamiliarity with the causal organism and rod-like morphology may delay diagnosis and treatment.

CONCLUSION: Gram-negative coverage should be considered in high-risk populations with cellulitis to prevent progression to fulminant abscesses, suppurative tenosynovitis and osteomyelitis.

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1. Introduction

Neisseria elongata is a rarely pathogenic microorganism. The *Neisseria* genus is composed of ten species, with only two considered as classically pathogenic to humans, *N. gonorrhoeae* and *N. meningitidis*. In 1990, the CDC received several strains of *N. elongata* originating from cases of bacterial endocarditis. The non-motile, gram-negative bacillus is typically found in the oral cavity and pharynx [1]. The *Neisseria* species is characterized as catalase-positive diplococci. The morphology of *N. elongata* is unusual as it is described as a rod-shaped organism and is catalase-negative and superoxol-negative [2]. Reports have identified the organism originating in blood and wounds and associated with infections such as endocarditis and osteomyelitis. These reports defined *N. elongata* as the causal organism in these infections and also mentioned additional cases of severe clinical manifestations, including congestive heart failure and myocardial abscesses [3]. An additional case report examined *N. elongata* in a patient with VSD and septicemia [4]. Review of the cases of *N. elongata* sepsis reveals associations with precipitating events, including dental procedures, open fractures, and history of heart disease or procedural intervention [5,6].

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Here, we describe a patient presenting with *N. elongata* cultures in a case of digital osteomyelitis. This case reported is reported in line with the SCARE criteria [7].

2. Presentation of case

A 63-year-old male with peripheral vascular disease and lower extremity wounds presented to his vascular surgeon. His past medical history was significant for morbid obesity (BMI 35.23 kg/m), end stage renal disease on dialysis, type II diabetes mellitus (A1C 6.1), hypertension, remote smoking (quit 20 years ago), and severe peripheral neuropathy and vasculopathy. Patient also had previous amputations of his left middle finger and left second toe due to gangrenous infections. Incidental examination of his upper extremities revealed concurrent right-handed redness and pain worsening over the past three days. The pain started distally in the right thumb with a recent history of minor thumb trauma. The pain was aggravated by palpation and movement with no remitting factors. The patient also noticed swelling in his right thumb and described progression of redness to his forearm. The patient was referred to the emergency department (ED) for urgent evaluation.

Upon assessment in the ED, the physical exam showed erythema and induration to the right hand, extending into the forearm (Fig. 1). Patient experienced tenderness to palpation along the volar thumb and into the distal forearm volarly with non-viable soft tissue distal phalanx. There was no pain to palpation in the small finger for suspi-



Fig. 1. Right hand, pre-operative gross image and X-ray.

tion of horseshoe abscess. Sensorineural examination was limited due to neuropathy. His vital signs on admission were a temperature of 98.4 °F (36.9 °C), respiratory rate of 20, and blood pressure of 121/62, with a pulse of 79, SpO₂ was reported at 94%. Lab values showed leukocytosis, with a count of 12,100/mm³ (neutrophils elevated at 9.71). Patient's POCT glucose level was 118 mg/dL on assessment in the ED. Blood cultures taken at arrival were negative.

On radiographic examination, the distal phalanx of the thumb showed extensive osteolysis of the distal phalanx. Further radiographic imaging with CT scan revealed inflammation throughout the right hand, forearm and carpal tunnel, as well as extensive vascular calcifications throughout the right upper extremity. In the operating room, exploration by the plastic surgery team revealed destructive osteomyelitis of the thumb distal phalanx and frank purulence in the radial bursa, carpal tunnel spreading into Parona's Space without any extension into the flexor sheath of the small finger. Irrigation and debridement, and partial thumb amputation were performed. Wounds were left open with vessel loop applied on edges for progressive closure post-op.

Culture swabs, soft tissue and bone biopsies were taken for culture and sensitivity. Gram stain revealed scant growth of beta-lactamase positive gram-negative rods. Culture demonstrated *Neisseria elongata* from right hand tissue. Echocardiogram 2D limited (2D doppler-color flow) with contrast was performed and showed no evidence of a mass or vegetation. Ejection fraction was 45–50% and the report indicated mild mitral regurgitation, and mild to moderate tricuspid regurgitation with a severely dilated right ventricle. The echocardiogram could not rule out endocarditis. TEE (transesophageal echocardiogram) was considered but was not clinically indicated as the patient was not bacteremic. Infectious disease was consulted, and sensitivities were followed, and antibiotics were adjusted from empiric vancomycin, clindamycin and meropenem to ceftazidime. The antibiotics were continued for a period of 6 weeks.

At his two-week postoperative follow-up appointment, the patient was doing well. The wounds demonstrated appropriate healing progression and were clean and intact with no signs of infection.

3. Discussion

We report a case of pathogenic *Neisseria elongata* infection causing digital osteomyelitis confirmed by tissue sampling and culture revealing rare gram-negative rods and growth of *N. elongata*. Gross pathology showed gangrenous necrosis with acute

and chronic inflammation. We confirm that *N. elongata* can be a causative organism in osteomyelitis. Given our patient's history of poorly controlled diabetes, dialysis-dependency and multiple comorbidities, risk of infection and associated decompensation was significant. Due to his vascular disease and diabetic neuropathy, minor trauma causing slow-to-heal wounds was more likely, as seen in the patient's lower extremity wounds and history of left-handed osteomyelitis and subsequent fingertip amputations.

This case of thumb distal phalanx osteomyelitis progressing to flexor tenosynovitis in the thumb and communication with Parona's Space follows the infection progression of a horseshoe abscess. This uncommon upper extremity condition involving the flexor tendon sheaths and bursae can spread from the first to fifth digits through the synovial sheath. Infection, similar to our case, spreads through Parona's Space and can lead to complications if left untreated such as neural compression, or carpal tunnel syndrome. Common etiologies include methicillin sensitive staphylococcus aureus, as well as MRSA in patients with a history of intravenous drug use, institutionalization or prior infection.

There have only been 23 documented cases involving pathogenic *N. elongata*, 21 of which were infectious endocarditis. While previous literature states the aggressive gram-negative rod has also caused chronic osteomyelitis, there are only two cases which were reported dating back to 1986 [8]. The first case of osteomyelitis describes a patient with multiple tooth extractions and delayed healing. *N. elongata* was identified 11 months following the index procedure of debridement of infected right maxillary tissue. The second case was identified in a patient with an open fracture of the leg, requiring multiple drainages over several years. Debridement recovered *N. elongata* from the right thigh. Limited information is available on the cases due to the early publication dates.

In clinical settings, unfamiliarity with the causal organism and rod-like morphology may delay diagnosis and treatment [9]. The CDC details biochemical tests performed to aid in the recognition of causative *Neisseria* organisms. The gram-negative rod can be identified using the Enzyme Substrate Test, as it produces no detectable acid from carbohydrates. The tube test utilizes the production of three enzymes, beta-galactosidase, gamma-glutamylaminopeptidase and hydroxyproplylaminopeptidase. Isolates are tested on a selective media for *Neisseria gonorrhoeae*, modified Thayer-Martin or equivalent. Supplemental tests are performed for species positive for hydroxyproplylaminopeptidase, such as *N. elongata* that may be confused for *N. gonorrhoeae* which is also hydroxyproplylaminopeptidase positive

[10]. CDC guidelines for differentiation of various *Neisseria* species allows for correct identification, especially of those species with either medicolegal implications or unusual morphologies as seen with this case of *N. elongata*.

The literature of pathogenic *N. elongata* further suggests that while cases are rare, prophylactic treatment should be initiated in susceptible patient groups. These patients include those with congenital or valvular heart disease, especially in the setting of dental procedures, as *N. elongata* exists in the normal oropharyngeal flora [11]. Additionally, as seen in our case, *N. elongata*, can be pathogenic in acute hand infections and implicated as a cause of acute osteomyelitis. Broad spectrum antibiotics with gram negative coverage should be considered in cases of high-risk populations with cellulitis in order to prevent progression to fulminant abscesses, suppurative tenosynovitis and osteomyelitis. Antibiotic therapy for previous *N. elongata* cases required the use of multi-drug regimens. One 2003 report summarizes the antibiotic treatments used, including the described endocarditis case utilizing 7 weeks of 6 mg of Ceftazidime, 0.5 MIC, as well as gentamicin [11].

4. Conclusion

Limited reports and unfamiliarity with the causal organism will continue to delay diagnosis and treatment of *Neisseria elongata* infections. This case is supportive evidence for consideration of gram-negative coverage in cases of high-risk populations with cellulitis in order to prevent progression to fulminant abscesses, suppurative tenosynovitis and osteomyelitis. Management of severe cases requires irrigation and surgical debridement, which can include amputation of the digits and extremities. Reports of recent cases identifying *N. elongata* in infectious conditions such as endocarditis, septicemia, and osteomyelitis will continue to enable better identification and treatment to address severe disease and prevent additional complications.

Declaration of Competing Interest

None.

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None.

Ethical approval

This study is exempt from ethical approval at this institution.

Consent

Written informed consent was obtained from the patient for publication of this case report and accompanying images. A copy of the written consent is available for review by the Editor-in-Chief of this journal on request.

Author contribution

A. Spielman and A. Ghumman drafted the manuscript. A. Ghumman and A. Lessard were involved in treatment of the patient. All authors review and approved manuscript for publication.

Guarantor

A. Lessard.

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