



## Case report

# Primary Actinomycosis in the breast caused by *Actinomyces neuui*. A report of 2 cases



B.S. Leenstra\*, C.C.M. Schaap, M. Bessems, N.H.M. Renders, K. Bosscha

Jeroen Bosch Ziekenhuis, Den Bosch, The Netherlands

## A B S T R A C T

Actinomycosis is a slowly progressive infection caused by anaerobic bacteria, primarily from the genus *Actinomyces*. Primary actinomycosis of the breast is rare and presents as a mass like density which can mimic malignancy. Mammography, ultrasonography and histopathologic examination is required for diagnosis. Treatment should consist of high doses of antibacterials for a prolonged period of time and possibly surgical drainage. Primary actinomycosis infections are commonly caused by *A. israelii*. *Actinomyces neuui* is a less common cause of classical actinomycosis. We present two cases of primary actinomycosis of the breast in two female patients caused by *A. neuui*.

## Introduction

Bacteria of the genus *Actinomyces* are gram-positive bacilli which generally colonize mouth, colon and vagina. These usually anaerobic bacteria uncommonly cause significant infections [1,2]. However, if actinomycosis occurs, it often presents as an indolent, slowly progressive infection characterized by abscess formation, which can progress across tissue boundaries, can present with mass like features which simulate malignancy, can develop into a sinus tract and/or develop clinically into a refractory or relapsing infection after a short course of therapy [2]. Actinomycosis is most commonly caused by *Actinomyces israelii* [3]. The species *Actinomyces neuui* is lesser known to cause classical actinomycosis [4]. Infections occur in the form of skin infections, endophthalmitis and endocarditis. Reports on *A. neuui* infections causing primary actinomycosis have occurred. To the authors knowledge only three cases have been reported [5–7]. We describe two cases of primary actinomycosis of the breast caused by *Actinomyces neuui*.

## Cases

A 43-year old woman with no significant medical history presented with complaints of an unilateral slowly growing painful mass in the left breast. No fever or symptoms of infection were present and laboratory findings were normal. Mammography (Fig. 1) and ultrasonography were conducted and showed a small abscess-like density retroareolarly.

A biopsy was obtained. Gram stain showed some leukocytes and

many gram-positive rods. Species identification was performed using matrix-assisted laser desorption ionization-time-of-flight mass spectrometry (MALDI-TOF MS; Bruker).

The isolate was identified as *A. neuui* (score value 2.368). Susceptibility testing for *A. neuui* was performed using E-tests (bioMérieux). At the time that culture findings became available, clinical symptoms improved. Therefore, the cultured *Actinomyces* was considered as skin commensals and a wait and see policy was installed. After several months, the patient returned to the outward clinic with complaints of pain in the breast. Physical examination of the breast showed mild erythema and ultrasonography showed a minimal increase of the lesion. The abscess was surgically drained and new purulent fluid from the abscess was sent to the laboratory. The material was processed as described above. After two days colonies were visible on the anaerobically incubated Schaedler agar. In contrast to initial culture findings, determination by the MALDI-TOF MS showed *Peptostreptococcus magnus* (score value 2.056). After four days there was also growth observed in the Brewer thioglycollate medium. Again *A. neuui* was identified. The isolate was shown to be susceptible to penicillin, amoxicillin, clindamycin, meropenem and vancomycin. Histopathological examination of the obtained tissue showed abscess formation in an infection with a sinus tract. Subsequently, the patient was treated for six weeks of intravenous penicillin followed by six months of oral amoxicillin successfully.

Secondly, a 73-year old woman with a history of bilateral breast cancer was seen at with severe pain and swelling of the left breast ten days after her ablation which was complicated by a large hamatoma

\* Corresponding author at: Henri Dunantstraat 1, 5223 GZ, Den Bosch, The Netherlands.

E-mail addresses: [b.leenstra@jzbz.nl](mailto:b.leenstra@jzbz.nl) (B.S. Leenstra), [Ch.Schaap@jzbz.nl](mailto:Ch.Schaap@jzbz.nl) (C.C.M. Schaap), [ma.bessems@jzbz.nl](mailto:ma.bessems@jzbz.nl) (M. Bessems), [N.Renders@jzbz.nl](mailto:N.Renders@jzbz.nl) (N.H.M. Renders), [K.Bosscha@jzbz.nl](mailto:K.Bosscha@jzbz.nl) (K. Bosscha).

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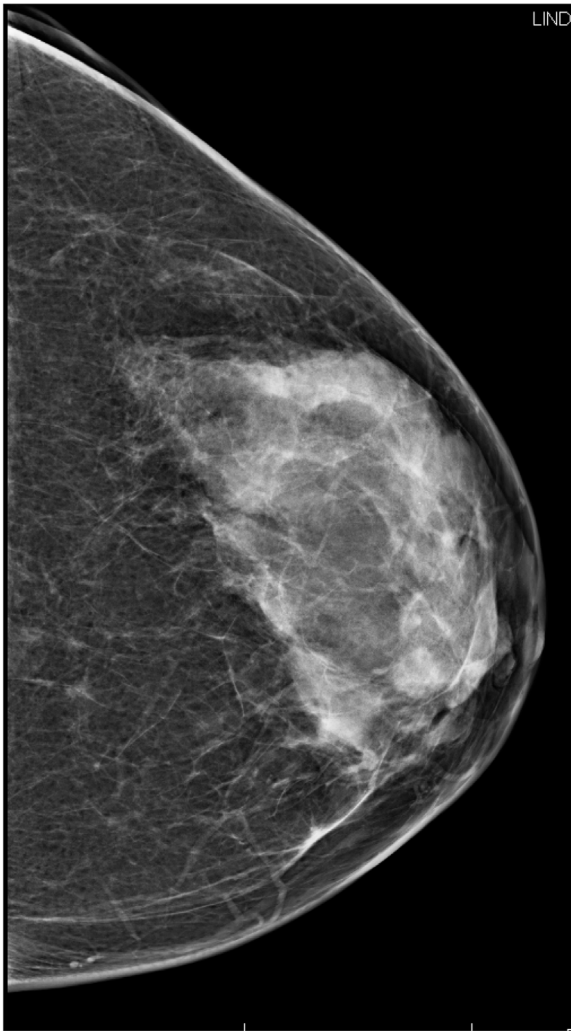


Fig. 1. Mammography of the left breast showing a confined density in the retroareolar area.

treated conservatively. Physical examination showed a painful swollen incision site with surrounding skin erythema and blistering. Laboratory findings showed a CRP level of 224. Ultrasonography (Fig. 2) showed a sharply defined inhomogeneous collection. A drain was inserted and fluid was obtained for analysis. Gram stain showed many leukocytes, some Gram-positive cocci and many Gram-positive rods. Determination using MALDI-TOF MS identified a *Staphylococcus aureus*. After 2 days there were colonies visible on the anaerobically incubated Schaedler agar. Determination by the MALDI-TOF MS revealed an *A. neuii*. Susceptibility testing was performed using E-tests. The isolate was susceptible to penicillin, amoxicillin, clindamycin, meropenem and vancomycin. Patient was successfully treated with 12 weeks amoxicillin orally. Unfortunately, patient returned four days after discharge with a relapse of the hematoma which was surgically evacuated. A month later at the outpatient clinic she showed full recovery.

## Discussion

Actinomycosis is infrequently seen. Approximately 80% of the primary actinomycosis infections are caused by *A. israelii*. Other isolates that have been identified as pathogens causing primary breast actinomycosis include *Actinomyces viscosus*, *Actinomyces turicensis* and *Actinomyces radingae* [8]. *A. neuii* is found in 17% of all clinical *Actinomyces* isolates. It differs from the other *Actinomyces* species because of its aerobic growth and its microscopic morphology without branching [4]. We were able to retrieve only three former cases of *A. neuii* causing primary actinomycosis. In the first case, treatment with antibacterials was unsuccessful and the lesion had to be surgically removed [5]. In the second case, patient was successfully treated with amoxicillin for 21 days [6]. In the third case, patient showed full recovery after a three month course of doxycycline [7].

Actinomycosis, regardless of species, should be treated with high doses of antibacterials for a prolonged period of time although therapy needs to be individualized. Current guidelines recommend high dose of penicillin intravenously for two to six weeks, followed by oral therapy with penicillin or amoxicillin for 6 to 12 months [3]. Alternative antibacterial treatments include doxycycline, erythromycin or clindamycin. If actinomycosis presents with well-defined abscess then surgical drainage followed by long-term antibacterial therapy is indicated. We describe this combination of treatment in our cases. In the first case

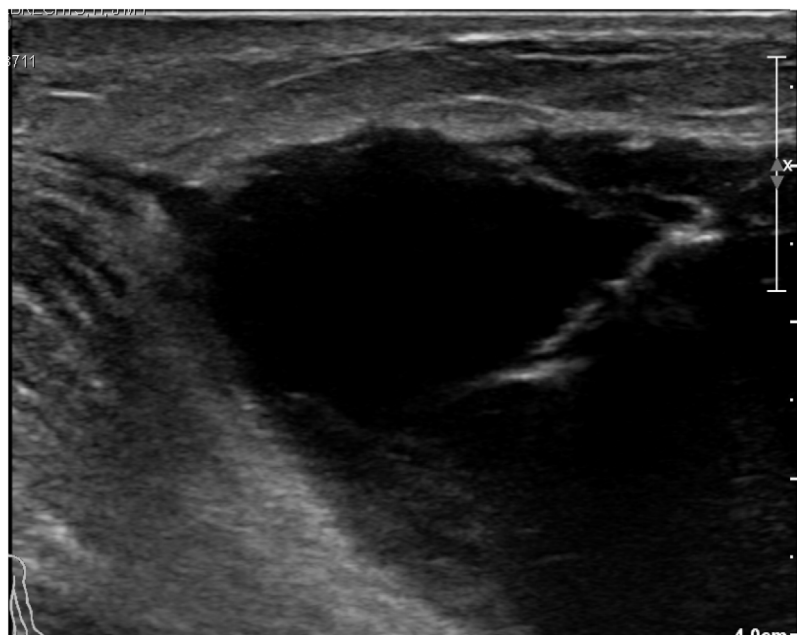


Fig. 2. Ultrasonography of the left breast showing drainage needle in infected hematoma.

initial cultures showed isolated *A. neuii*. Because signs of infection had decreased this finding was interpreted as skin flora and not as a pathogen. Therefore medical treatment was considered unnecessary.

In retrospect, if antibacterial therapy had been given at first presentation recurrence of the infection might have been prevented. *Actinomyces* species morphologically resemble diphtheroids and could be dismissed as skin commensals, even when isolated from an abscess sample. In both of our presented cases we found different organisms in the culture additional to *A. neuii*. In the first case we found *P. magnus* and in the second case *S. aureus* was present. Zelyas et al. also described *A. neuii* with mixed anaerobic organisms and skin flora [1]. Although *A. neuii* infection is known for its chronicity, the influence of these mixed organisms on the severity of infections remains unclear [1]. Further research on the effect of mixed organism on the severity of an *A. neuii* infection is required.

## References

- [1] Nathan, et al. Infections caused by *Actinomyces neuii*: a case series and review of an unusual bacterium. *Can J Infect Dis Med Microbiol* 2016;2016.
- [2] Smego Jr., Foglia. Actinomycosis. *Clin Infect Dis* 1998;26(6):1255–63.
- [3] Russo TA. Agents of actinomycosis. In: Mandell GL, Bennett JE, Dolin R, editors. *Mandel, Douglas and Bennett's principles and practice of infectious disease*. 4th ed. New York: Churchill Livingstone; 1995.
- [4] von Graevenitz A. Actinomyces neuii: review of an unusual infectious agent. *Infection* 2011;39:97. <http://dx.doi.org/10.1007/s15010-011-0088-6>.
- [5] Roustan, et al. Primary actinomycosis of the breast due to Actinomyces neuii. *J Gynecol Obstet Biol Reprod (Paris)* 2010;39(February (1)):64–7. <http://dx.doi.org/10.1016/j.jgyn.2009.09.005>. Epub 2009 Oct 12.
- [6] Lacoste C, et al. Breast *Actinomyces neuii* abscess simulating primary malignancy: a case diagnosed by fine-needle aspiration diagn cytopathology. *Diagn Cytopathol* 2009;37(April (4)):311–2. <http://dx.doi.org/10.1002/dc.21044>.
- [7] Olson JM, Vary Jr. J.C. Primary cutaneous Actinomyces neuii infection of the breast successfully treated with doxycycline. *Cutis* 2013;92(December (6)):E3–4.
- [8] Valour F, et al. Actinomycosis: etiology, clinical features, diagnosis, treatment, and management. *Infect Drug Resist* 2014;5(July (7)):183–97. <http://dx.doi.org/10.2147/idr.s39601>. eCollection.