CASE REPORTS



Primary Actinomycosis of the Foot in a 37-year-old female Patient: A Case Study and Review of the Literature.

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Abstract

A parasitic disease called actinomycosis occurs when bacteria spread through body tissues, causing abscesses, inflammation, and pain. It affects the skin or deeper parts of the body and can sometimes affect the blood.[1]

The symptoms of actinomycosis may mimic those of other diseases or even neoplasms. Remission and exacerbation of symptoms occurring in parallel sequence with the initiation and cessation of therapy is a phenomenon that should increase suspicion of actinomycosis in any of its manifestations. [2]

Microbiology, histopathology, and MRI revealed the presence of an unusual infectious agent, Actinomyces spp., which is also known as Madura foot [3] This implies a serious difficulty in getting a bactericidal concentration of the effective drug into areas of active infection and seriously questions the possibility of restoring the involved tissues [4, 8]

The diagnosis was confirmed by isolation of the organisms by anaerobic culture giving typical molar tooth colonies. Final confirmation was done by histopathological examination.

Case report; The patient's condition dates back 5 years before the operative treatment when for the first time the patient reported that she was stabbed with a foreign body in the area of the left foot. The patient was treated at home and in the beginning, did not go to the doctor. She occasionally complained of pain and swelling but the pain was not distressing, with no fever and no swelling of the foot...

Keywords: Actinomycosis, Foot, Unusual Wounds, Fistula, Amputation

Introduction:

Actinomycosis is an indolent, slowly progressive, suppurative infection caused by gram-positive branching bacteria of the genus Actinomyces. The organism is a member

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of the oral and gastrointestinal microflora of humans. The disease actinomycosis most commonly occurs in 3 body regions: cervicofacial (55% of patients), abdominopelvic (20%), and pulmonothoracic (15%).[2]

Involvement of other parts of the body is uncommon and usually secondary to a lesion in one or the other of the above sites. Extremity disease can occur by secondary involvement through direct extension or hematogenous spread. However, primary actinomycosis of an extremity is very rare

Case report

The patient's condition dates back 5 years before the operative treatment, when for the first time the patient reported that she was stabbed with a foreign body in the area of the left foot. (Fig.1)





The patient was treated at home and in the beginning did not go to the doctor. She occasionally complained of pain and swelling but the pain was not distressing, with no fever and no swelling of the foot.

She came to our clinic for the first time 2.5 years ago after the onset of symptoms. Pain and swelling became more intense and the performance of daily activities became more difficult.

The swelling of the foot increased, the color began to become easily liquefied and a yellowish-white secretion flowed from several formed fistulas through the foot, especially the plantar side.

During this period the patient is afebrile and in good condition, but with difficulty moving due to the more obvious

swelling of the whole foot, with slight fluid discoloration and present fistulas that rarely secrete, most of them on the plantar side and some on the dorsal side.

Before being admitted to our clinic, the patient was conservatively treated several times in the Clinic for Infectious Diseases and Febrile Conditions, Plastic and Reconstructive Surgery – Skopje. During the intensive antibiotic therapy, the swelling and pain subsided, but not completely.

After the admission at our clinic, all clinical and paraclinical diagnostic procedures were performed: MRI, CT of the left foot, bone scan with TC 99.

Laboratory examinations (relatively elevated inflammatory parameters), X-ray and ultrasonography of



Fig. 1. Patient With Actinomycosis Foot Infection With Multiple Sinuses



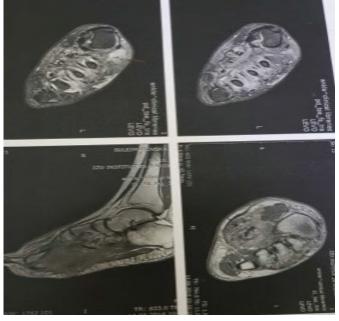


Figure 2. MRI of the left foot – DOT SIGNS



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the lungs and other examinations. (Fig. 2)

During the entire period of hospitalization, the patient was treated with combination of two antibiotic (Climycin and Ceftraxone i.v), antifungal drug (Diflazone) and topical treatment with rinsing and dressings of the foot.

An open biopsy was performed in the operating room, biopsy material was taken - altered tissue, yellowish gray-whitish granulation in front of the plantar pedis, and the material was sent for pathohistological analysis at the Institute of Pathology (PTH attached of Actinomycosis)

Given that this pathology is very rare in our country and also the location of actinomycosis in the foot is very rare (3-4%), the patient was explained in detail about the course of the disease and the outcome of the disease in the future, continuous antibiotic treatment and final decision for radical surgical treatment - amputation of the foot.

The patient went on her own initiative to a private

clinic where she was also treated with systemic and local antibiotics and at one time an incision was made, drainage and evacuation of abscesses and placed on vacuum treatment on the left foot. She was treated there for almost a year without improvement of the local finding of the foot and without improving the overall health and quality of life.

The patient was re-admitted to our ins due to deteriorating health, especially the local finding of the foot with the presence of huge swelling of the left foot, fluid discoloration and lost sensitivity of the toes of the left foot. Several fistulas were present but without excessive puss drainage.

The diagnostic procedures were re-performed with MRI, CT angiography, echo of the abdomen, X-ray of the lungs and a set of laboratory tests. After completing the above diagnostic procedures and with previous PTH analyzes (actinomycosis), the patient was offered surgery –

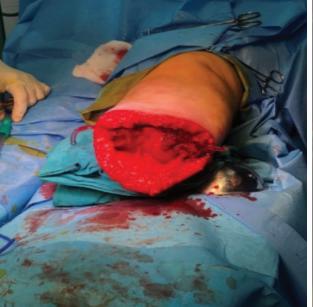




Figure 3. The procedure of amputation and the suture of the wound







Figure.4. RTG of the left foot and leg (A) before and (B) after amputation



below the knee amputation on the left side.

The operation was performed with prior anesthesiologic preparation. The operation was performed in spinal anesthesia, under tourniquet in the middle of the tibia. Fibula was resected circa 4 cm above the tibial cut. (Fig 3)

After the operation, the wound was closed layer by layer with previous ligation and hemostasis of blood vessels and with a good flap on the skin. A pathohistological analysis of the amputated foot was ordered and it was sent to the Institute of Pathology.

During the stay, a physiatrist and physiotherapist were also consulted for further advice on movement. An appropriate Center for orthotics was consulted for the processing of an appropriate carbon dynamic prosthesis, given the young age of the patient and the daily engagements.

Postoperatively, the patient is in good general health, a local wound treatment and dressing was done and with systemic antibiotic - Vancomycin Sutures removed on the 14th postoperative day.

The patient is discharged in good general and local condition. Rehabilitation has been performed and the patient has undergone a permanent knee prosthesis, which has changed the quality of life, daily (fig.4) engagements, movement and functionality without any medication.

Discussion:

Several Actinomyces species are endogenous to the body and are part of the human microflora. The pathogenic counterpart is parasitic and is spread when there is disruption in the mucosal barrier, allowing the organism to enter soft tissues where blood supply is impaired.[5]

The World Health Organization has recently recognized mycetoma as a neglected tropical disease caused by either filamentous fungi (eumycotic) or bacteria (actinomycotic). Mycetoma can be prevented and treated, especially in its early stages, but it has a high morbidity rate and a large economic impact [6] Environmental actinomycotic organisms can induce a dense fibrotic tissue response without fistulas. The result was a painful, sclerosing, nonsuppurative infection of the bone and surrounding tissues in our patient.[7]

Unlike other Actinomyces species, A. Meyeri frequently causes pneumonia and has a tendency for hematogenous dissemination. When penicillin is administered for several months and surgical procedures are performed, when necessary, the outcome for these patients is excellent. [8]

If the diagnosis is delayed, the affected part may have to be amputated. We present a case of histologically proven mycetoma with MRI features characteristic of the recently described "dot-in-circle" sign. [8] Despite the fact that biopsy and microbiological culture provide definitive diagnoses, these are often difficult to obtain. [9]

As a result of the breakdown of nodules and formation of discharging sinuses, bacterial infections may progress to full-blown bacteremia or septicemia, leading to death. Disfiguring sequelae result as well. [10]

Among the countries where mycetoma is studied rigorously, Mexico, India, and Sudan are the most common sites. There is discovered that mycetoma has spread to the United States, Venezuela, Italy, China, and Australia as well. Mycetoma is reported far outside of the tropics. In the Americas, bacterial forms predominate, but in Africa and Asia, the picture is more varied. [11]

A better understanding of mycetoma epidemiology will enable more education, preventive measures, and treatment to be directed to at-risk areas, helping to reduce disease burdens. [12, 13]

Conclusion:

As the disease is relatively slow and painless, patients don't report it early, and therefore it is diagnosed at a late stage. The earlier the disease is reported to the primary care physician, and the sooner the disease is diagnosed, the lower the likelihood of morbidity. While working in fields, it is also important to wear appropriate footwear.

Declaration of any potential financial and nonfinancial conflicts of interest:

The authors declare that they have no known competing financial interests or personal relationships that could have appeared to influence the work reported in this paper.

Ethics approval and consent to participate – all the patients/their parents have signed informed consent.

Consent for photography- all the patients/their parents have signed informed consent

Consent for publication - all the patients/their parents have signed informed consent

Clinical trial registration information provided – not applicable

We confirm that the manuscript, including related data, figures and tables has not been previously published and that the manuscript is not under consideration elsewhere.

References:

- Spilsbury B.W., Johnstone F.R.C. The clinical course of actinomycotic infections: a report of 14 cases. Can J Surg. 1962; 5:33–48
- Bennhoff DF. Actinomycosis: diagnostic and therapeutic considerations and a review of 32 cases. Laryngoscope. 1984 Sep;94(9):1198-217. doi: 10.1288/00005537-198409000-00013. PMID: 6381942.
- Al Gannass A. Chronic Madura foot: mycetoma and/ or Actinomyces spp or actinomycosis. BMJ Case Rep. 2018; bcr2018224859. Published 2018 May 22. doi:10.1136/bcr-2018-224859
- Harvey J., Cantrell J., Fisher A. Actinomycosis: Its recognition and treatment. *Ann Intern Med.* 1957; 46:868– 885



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 Bettesworth J, Gill K, Shah J. Primary actinomycosis of the foot: a case report and literature review. J Am Col Certif Wound Spec. 2009 Jul 3;1(3):95-100. doi: 10.1016/j. jcws.2009.06.004. PMID: 24527123; PMCID: PMC3478921.

- Verma P, Jha A. Mycetoma: reviewing a neglected disease. Clin Exp Dermatol. 2019 Mar;44(2):123-129. doi: 10.1111/ ced.13642. Epub 2018 May 28. PMID: 29808607.
- 7. Vandevelde AG, Jenkins SG, Hardy PR. Sclerosing osteomyelitis and Actinomyces naeslundii infection of surrounding tissues. Clin Infect Dis. 1995 Apr; 20(4):1037-9. doi: 10.1093/clinids/20.4.1037. PMID: 7795046.
- 8. Iniesta A, Baptista C, Guinard D, Legré R, Gay A. Mycétome actino-mycosique du pied, à propos d'un cas [Mycetoma of the foot: a case report]. Ann Chir Plast Esthet. 2015 Apr;60(2):164-7. French. doi: 10.1016/j.anplas.2013.11.002. Epub 2013 Dec 28. PMID: 24380724.
- Apothéloz C, Regamey C. Disseminated infection due to Actinomyces meyeri: case report and review. Clin Infect Dis.

- 1996 Apr;22(4):621-5. doi: 10.1093/clinids/22.4.621. PMID: 8729199.
- Javed F, Nazir R, Sharma M, Yasir S, Babar S. Dot-in-Circle Sign - A Diagnostic MRI Sign for "Madura Foot". J Coll Physicians Surg Pak. 2017 Mar;27(3): S8-S10. PMID: 28302229.
- 11. Kumar J, Kumar A, Sethy P, Gupta S. The dot-in-circle sign of mycetoma on MRI. Diagn Interv Radiol. 2007 Dec;13(4):193-5. PMID: 18092291.
- 12. Venkatswami S, Sankarasubramanian A, Subramanyam S. The madura foot: looking deep. Int J Low Extrem Wounds. 2012 Mar;11(1):31-42. doi: 10.1177/1534734612438549. Epub 2012 Feb 14. PMID: 22334597.
- Emery D, Denning DW. The global distribution of actinomycetoma and eumycetoma. PLoS Negl Trop Dis. 2020 Sep 24;14(9): e0008397. doi: 10.1371/journal.pntd.0008397. PMID: 32970667; PMCID: PMC7514014.

