



Primary actinomycosis of the breast in postmenopausal women



Ann Ital Chir, Digital Edition 2019, 8
pii: S2239253X19031232 - Epub, December 20
free reading: www.annitalchir.com

Orçun Yalav, Uğur Topal, Ayşe Gizem Ünal, Kubilay Dalci

Department of General Surgery, Cukurova University Faculty of Medicine, Cukurova/Adana, Turkey

Primary actinomycosis of the breast in postmenopausal women

INTRODUCTION. *Actinomycosis is a chronic infection caused by actinomyces species characterized by an abscess formation, tissue fibrosis, and draining sinuses. Primary actinomycosis of the breast is rare.*

PATIENTS AND METHODS: *In this paper we present a 64-year-old postmenopausal woman. For the diagnosis of primary actinomycosis of the breast, mammography, ultrasonography, MRI, and histopathologic examinations are required. Microbiological culture and histopathology are of the most importance during the process of diagnosis. In our case, at the intersection of the sternum and the lower inner quadrant of the right breast, there was a 1 cm wide fistula opening, and an abscess. A. israelii has been isolated from the microbiological culture taken from the lesion*

RESULTS: *An optimal surgical resection of infected tissues has been performed as the treatment with the wound left open for tertiary healing. The patient was given sulbactam 4*1 gr/day intravenously for 4 weeks post-op.. Recurrence was not detected during the yearly follow up procedures*

CONCLUSIONS: *Actinomycosis should be considered when differentially diagnosing clinical instances of suppurative or granulomatous infections of the breast and mass regions that can't be ruled out as malignancies. Early diagnosis will save the patient from unnecessary surgical operations and ineffective antibiotic treatments.*

KEY WORDS: Actinomycosis, Breast abscess, Breast diseases

Introduction

Actinomycosis is a rare slow-spreading chronic disease, characterized by abscess formation, tissue fibrosis and draining sinus infections. Actinomycosis is caused by actinomyces like organisms, which are Gram-positive, anaerobic or microaerophilic bacteria that shows filamentous growth and branching. The most frequent cause is *A. israelii*, which is present in 78% of patients ¹.

Actinomyces like organisms can be found in oropharynx, gastrointestinal tract and the flora of the female genital tract. They can cause opportunistic infections on these

mucosal surfaces in cases of tissue damage of any cause. Since these microorganisms are hard to observe microscopically, and hard to produce in culture, they are usually diagnosed late, or are completely misdiagnosed ^{2,3}.

Along with clinical findings, microbiological and histopathological examinations assist the process of diagnosis. Primary actinomycosis of the breast in postmenopausal women is quite rare and in medical literature it was first described by Ammentorp in 1893 ⁴.

In medical literature, there has been around 30 reported cases of primary actinomycosis of the breast ⁵. This work was reported in line with the Surgical Case Report Guidelines (SCARE) criteria ⁶.

CASE PRESENTATION

The patient is a 64-year-old Turkish woman who was admitted to our clinic of infectious diseases with discharge from the right nipple and a high body temperature. The patient was admitted to the infectious diseases ward,

Pervenuto in Redazione Giugno 2019. Accettato per la pubblicazione Ottobre 2019

Correspondence to: Uğur Topal M.D, Department of General Surgery, Cukurova University Faculty of Medicine, 01100 Sarıçam/Adana, Turkey (e-mail: sutopal2005@hotmail.com)



Fig. 1: MRI was showed . lesion enveloping the pectoral muscle from the midline intersection of both pectoral muscles, lengthening into the right breast, 1*5cm with a peripheral contrast area.

imipenem 4*500mg + daptomycin 1*350mg was started, she took it for 18 days, and was discharged upon decrease in nipple discharge. In the first month after the initial discharge, the patient developed a fistulizing lesion under the right nipple and applied to the general surgery department.

She is a nonsmoker patient and was diagnosed with coronary artery disease, type 2 Diabetes Mellitus. She had a history of cholecystectomy and no history of lung disease, breast trauma, gingivitis or tooth problems. She was using acetylsalicylic acid 1*100 mg and Pioglitazone 2*1gr.

During the physical assessment, her blood pressure level was 120/70 mmHg, respiratory rate was 20, Pulse was 80/min, body temperature was 37.4°C Her body mass index was 28.9 kg/m². Her general condition was normal and the patient was conscious and cooperative. At the intersection of the sternum and the lower inner quadrant of the right breast, there was a 1 cm wide fistula opening. A clinical breast examination yielded a normal result for both breasts and axilla.

The patient's blood and biochemical values were in a normal range, and blood cultures were negative. A chest x-ray was normal. A mammography showed a liposclerous breast pattern. A sonogram of the breast reported a 3*1 cm wide lesion that dissects subcutaneous fat tissue, and surrounding tissues on the lower inner quadrant of the right breast. The lesion was in accordance with an organizing abscess formation. In the dynamic breast MR of the patient, a lesion enveloping the pectoral muscle from the midline intersection of both pectoral muscles, at the lower half of the bilateral medial area, lengthening into the breast, 1*5cm in the left and a shorter line in the right, with a peripheral contrast area (Fig. 1).

The patient was operated with a diagnosis of fistulization of the breast. The operation was performed under general anesthesia in supine position. An experienced team of breast surgeons performed the operation. During the exploration the fistule tract was found to be extending from the side of sternum through the right axilla, and the abscesses and the fibrous tissue were drained and resected. (Fig. 2).

The whole fistula tract was excised. Pathologic examination of a 10*5*4 cm tissue specimen Duration of the operation was 1 h 15 min and blood loss was 20 ml. The defect was followed up for 7 days with open wound dressing. Vacuum assisted closure (VAC) (Kinetic Concepts Inc., San Antonio, TX) was applied to the patient for 3 sessions. The wound was sutured after sufficient granulation (Fig. 3).

Histopathologic examination showed active chronic suppurative fistulized inflammation granulation tissue. The swab culture taken from the wound showed *A israelii* reproduction.

The patient was given sulbactam 4*1 gr/day intravenously for 4 weeks, and a treatment plan of 2*1 gr/day oral amoxicillin-clavulanate was prescribed for the first 12 months after surgery. The patient had no specific post-operative complications. and the patient was discharged 30 days post op.

The patient didn't experience any follow up problems after the 1st month. During the first 12 months of fol-



Fig. 2: The fistule tract was found to be extending from the side of sternum through the right axilla, and the abscesses and the fibrous tissue were drained and resected.



Fig. 3: The wound sufficient granulation after VAC.

low-up process, there has been no clinical symptom of a breast infection and the patient is still attending the regular follow-up controls. The patient should be closely followed for possible breast infections. Written informed consent was provided by the patient.

Discussion

Actinomyces are Gram-positive anaerobic or microaerophilic bacteria that show filamentous growth and branching. They could be found in oropharynx, gastrointestinal tract and the normal flora of female genital tract. Therefore, 60% of the infections are found around the cervicofacial area, 22% around abdominal area and 15% around the thorax ⁷.

They can mimic conditions such as malignancy or tuberculosis during diagnosis. Primary breast actinomycosis is more uncommon than these disorders. The most frequent cause of primary breast actinomycosis is *A. israelii*, present in 78% of patients ¹ along with *A. israelii*, other actinomyces organisms such as *A. turicensis*, *A. radingae*, *A. Neuii*, *A. Odontolyticus*, *A. europaeus*, *Actinomyces meyerii*. Those species has been isolated in primal breast actinomycosis some cases ⁵.

Risk factors for development of primary breast actinomycosis include smoking, diabetes, presents as a chronic, recurrent abscess and fistules Breast actinomycosis is primary when inoculation occurs through the nipple. Secondary actinomycosis of the breast refers to the extension of a pulmonary infection through the thoracic cage in a process that can affect the ribs, muscles and finally the ⁸. Our breast actinomycosis patient didn't have any pulmonary infections. In mammary actinomycosis, masses with uneven edges or asymmetric density, thickening of the skin, and thick-walled collection (abscess) behind the nipple or collection areas showing fistulization to the skin can be observed in mammography ⁹.

In our patient, abscess and fistula tract were present in the USG. Imaging methods like MRI and CT are used to determine whether the malignancy originates from retraction or inflammation. This differential diagnosis method informs us a great deal. Actinomycosis could be diagnosed by observing characteristic sulfur granules during histopathological examination. These granules are composed of basophilic structures with elongated eosinophilic clubs radiating from the periphery. The granules contain gram-positive, non-acid-fast bacteria. New methods of identification, such as matrix-assisted laser desorption ionization-time of flight mass spectrometry (MALDI-TOF MS), allow rapid and reliable identification of many bacteria, including actinomyces like organisms ¹⁰.

Primary breast actinomycosis patients should go through an extensive antibiotic treatment after draining of all abscesses and surgical removal of all fistules and sinuses. Common treatment regimens consist of high-dose penicillin intravenously for 2 to 6 weeks, followed by an oral therapy with penicillin or amoxicillin for 6 to 12 months. In our case we prescribed our patient sulbactam for the first 4 weeks and an amoxicillin-clavulanate treatment for the next 12 months.

Conclusion

In conclusion, actinomycosis is a rare chronic disease caused by actinomyces spp., these anaerobic Gram-positive bacteria form a disease that can come up in different parts of the body with different pathologies. Actinomyces-associated breast infections are problematic, difficult to diagnose, difficult to treat and often missed by experienced clinicians. Actinomycosis should be considered when differentially diagnosing clinical instances of suppurative or granulomatous infections of the breast and mass lesions that can't be ruled out as malignancies. Early diagnosis will save the patient from unnecessary surgical operations and ineffective antibiotic treatments.

Riassunto

L'actinomicosi è un'infezione cronica causata da specie di actinomyces caratterizzata da formazione di ascessi, fibrosi tissutale e fistolizzazione, di rara localizzazione al seno.

Viene qui presentato il caso di una donna di 64 anni in postmenopausa. La diagnosi di actinomicosi primaria del seno ha richiesto esami mammografici, ecografici e con RM e controlli istopatologici. La coltura microbiologica e l'istopatologia sono della massima importanza durante il processo di diagnosi. Nel nostro caso, all'intersezione dello sterno e del quadrante interno inferiore del seno destro, c'era una fistola larga 1 cm e un asces-

so. Dalla coltura microbiologica dall'essudato fistoloso è stato isolato *Actinomyces israelii*.

È stata eseguita una resezione chirurgica ottimale dei tessuti infetti, lasciando la ferita aperta per una guarigione per terza intenzione. Per 4 settimane dopo l'intervento è stato somministrato Sulbactam per via endovenosa (1 gr x 4/die). Ad un anno non si riscontrata alcuna recidiva.

In conclusione l'actinomicosi deve essere presa in considerazione nella diagnosi differenziata anche in caso di infezioni suppurative o granulomatoze del seno e delle masse di tessuti che non possono essere esclusi per natura neoplastica. La diagnosi precoce salverà il paziente da interventi chirurgici inutilmente demolitivi e da trattamenti antibiotici inefficaci.

References

1. Lerner PI: *Actinomicose*. In: Harrison TR, Ed. *Medicina interna*. Rio de Janeiro: McGraw-Hill, 1995; 732-35.
2. Kaya D, Demirzen Ş, Beksac MS: *Actinomyces türlerinin*. *Biyolojik Özellikleri Biyoloji Bilimleri Araştırma Dergisi* 2 (2): 25-28, 2009.
3. Guglielmi A, Veraldi GF, Negri A, Battocchia A: *Primary hepatic actinomycosis: A clinical case and review of the literature*. *Ann Ital Chir*, 1991; 62:185/9.
4. Lloyd-Davies JA: *Primary actinomycosis of the breast*. *Br J Surg*, 1951; 38:378-81.
5. ABing AU, Loh SF, Morris T, Hughes , Dixon MJ, Helgason O: *A actinomyces species isolated from breast infections*. *Journal of clinical microbiology*, 2015; 53(10):3247-255.
6. Agha RA Fowler AJ, Saeta Barai I, Rajmohan S, Orgill DP: SCARE group: *The SCARE statement: Consensus-based surgical case report guidelines*. *Int J Surg*, 2016; 34: 180-86.
7. Palonta F, et al.: *Actinomycosis of the masseter muscle: Report of a case and review of the literature*. *J Craniofac Surg*, 2003; 14:91.
8. Salmasi A, Asgari M, Khodadadi N, Rezaee A: *Primary actinomycosis of the breast presenting as a Breast Mass*. *Breast Care*, 2010; 5:105-07.
9. Çiçek-Sentürk G, Altay FA, Köybasioglu F, Ulu-Kiliç A, Öztürk, E, Albayrak, Y, Sencan I: *Two different actinomycosis cases presented with mass, prediagnosed as malignancy in breast and scrotum*. *Klinik Dergisi*, 2013; 26(3):122.
10. SchmittBH, Scott Cunningham AAL, Dailey DR, Gustafson RP: *Identification of anaerobic bacteria by bruker biotyper matrix-assisted laser desorption ionization-time of flight mass spectrometry with onplate formic acid preparation*. *J Clin Microbiol*, 2013; 51:782-86.