

CASE REPORT

Multiple pseudotumoral lesions of the abdominal wall caused by *Actinomyces* Case ReportLesiones pseudotumorales múltiples de la pared abdominal por *Actinomyces*. Presentación de un casoMúltiplas lesões pseudotumorais da parede abdominal por *Actinomyces*. Apresentação de um casoIlian Esteban Tarife Romero^{1*} , Leyanis González Baigorriá¹ , YanderLuisIzaguirreCampillo^{II} ¹Facultad de Ciencias Médicas Mayabeque. Mayabeque, Cuba.^{II}Universidad de Ciencias Médicas de Ciego de Ávila. Ciego de Ávila, Cuba.* Corresponding author: itarife6@gmail.com

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ABSTRACT

Actinomyces is a family of acid-fast, non-sporulated Gram-positive bacilli that constitute a diverse collection of bacteria that colonize the skin and mucosal surfaces. Isolation and identification occurs only in the minority of cases. The objective of the article is to describe a case with pseudotumoral lesions of the abdominal wall caused by *Actinomyces*. This is a 54-year-old white female patient who presents with non-specific and local symptoms such as a visible and palpable tumor mass, painful in the right upper quadrant and epigastrium, diagnosed with severe chronic panniculitis with granulation and *Actinomyces* infection. Surgical treatment was performed. The location of colonies in the abdominal wall is not frequent and a certain diagnosis is made by obtaining a surgical biopsy and observing histological slides.

Keywords: *Actinomyces*; actinomycosis; abdominal neoplasms

RESUMEN

Actinomyces es una familia de bacilos Gram positivos no esporulados ácido-resistentes que constituyen una colección diversa de bacterias que colonizan la piel y las superficies mucosas. El aislamiento e identificación ocurre solo en la minoría de los casos. El objetivo del artículo es describir un caso con lesiones pseudotumorales de la pared abdominal producidas por *Actinomyces*. Se trata de una paciente femenina, blanca, de 54 años de edad, que acudió con síntomas inespecíficos y locales como masa tumoral visible y palpable, dolorosa en hipocondrio derecho y epigastrio, se diagnosticó de paniculitis crónica severa con granulación e infección por *Actinomyces*. Se realizó tratamiento quirúrgico. La localización de las colonias en la pared abdominal no es frecuente y el diagnóstico de certeza se tiene por obtención de biopsia quirúrgica y observación en láminas histológicas.

Palabras clave: *Actinomyces*; actinomicosis; tumores abdominales



RESUMO

Actinomyces é uma família de bacilos Gram-positivos não esporulados, álcool-ácido resistentes, que constituem uma coleção diversificada de bactérias que colonizam a pele e as superfícies mucosas. O isolamento e a identificação ocorrem apenas na minoria dos casos. O objetivo do artigo é descrever um caso de lesões pseudotumorais da parede abdominal causadas por *Actinomyces*. Paciente do sexo feminino, 54 anos, branca, que apresenta sintomas inespecíficos e locais como massa tumoral visível e palpável, dolorosa em

quadrante superior direito e epigástrio, com diagnóstico de paniculite crônica grave com granulação e infecção por *Actinomyces*. Foi realizado tratamento cirúrgico. A localização de colônias na parede abdominal não é frequente e o diagnóstico certo é feito pela obtenção de biópsia cirúrgica e observação de lâminas histológicas.

Palavras-chave: Actinomyces; actinomicose; neoplasias abdominais

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INTRODUCTION

Actinomyces is a family of Gram-positive, non-sporulating acid-fast bacilli that constitute a diverse collection of bacteria that colonize skin and mucosal surfaces. They grow slowly in cultures under anaerobic conditions. Thirteen species are known, of which *A. israelii*, *A. naeslundii*, *A. viscosus*, *A. odontolyticus*, *A. pyogenes* and *A. meyeri* cause disease in humans, however, it is *A. israelii* that causes the most disease.⁽¹⁾

The most common sites of infection are the cervicofacial region, thoracic, abdominopelvic and central nervous system. Long-term use of an intrauterine device (IUD) can, in 3% of cases, promote the development of actinomycosis in the female genital tract and leads to the formation of abscesses with non-specific symptoms.^(1,2)

Such infections often spread to the surrounding tissues by direct extension and form fistulas that in some cases lead directly to the skin. There is formation of abundant dense granulomatous and fibrous tissue. Symptoms include fever, pain, weight loss, swelling at the site of infection and abscesses. In most cases the diagnosis is very difficult and is obtained after surgery or by microbiological and pathological findings which are the methods of certainty.^(1,3)

Infections are infrequent due to their low prevalence, with atypical clinical manifestations, especially in cases that present as tumor lesions, infiltrating soft tissues or adjacent organs. It is characterised as a chronic, suppurative, granulomatous, non-contagious, slowly progressive disease. It is not age group specific, however, it is thought to be common in middle-aged people and less common in individuals under 10 and over 60 years of age.^(4,5)



Bacteriological identification of *Actinomyces* from a sterile site confirms the diagnosis of actinomycosis, but isolation and identification occurs only in a minority of cases due to prior antibiotic therapy, inadequate culture conditions and contamination by other microorganisms. Appropriate clinical specimens are tissues from surgical biopsies. If actinomycosis is suspected, the microbiologist should be consulted to ensure proper culture management.⁽⁶⁾

We report an actinomycosis of the abdominal wall, a rare entity with an uncommon diagnosis. The description facilitates understanding of the clinical features and establishes a reference point for treating future patients. This article aims to describe a case with multiple pseudotumoral *Actinomyces* lesions of the abdominal wall.

CASE PRESENTATION

Female patient, white, 54 years of age, with a personal pathological history of compensated arterial hypertension (AHT), on regular treatment with enalapril (20 mg) and hydrochlorothiazide (25 mg/day). Family history of ischaemic heart disease (mother deceased), she reported treatment for dental phlegmon with the stomatology service and use of an intrauterine device (IUD) placed more than seven years ago. She denied recent abdominal trauma, drug allergy or other relevant information for the case.

She attended the General Surgery Ward of the "Aleida Fernández Chardiet" Teaching-Clinical-Surgical Hospital in the municipality of Güines, in the province of Mayabeque, Cuba, where she reported general malaise and malaise, accompanied by diffuse abdominal pain of eight days' evolution that began spontaneously in the left upper hemiabdomen and hypogastrium area with no other irradiation. Occasionally it behaved like cramps without relief when changing position or taking non-steroidal anti-inflammatory drugs; he also reported the appearance of a visible and palpable mass 72 hours after the onset of the first symptoms, in the same location, accompanied by a fever of 39 to 40 °C and abundant vomiting of food content, the last of which was very dark in colour.

Physical examination revealed low-coloured mucous membranes, polypnea (respiratory rate of 36 breaths per minute), tachycardia (heart rate of 125 beats per minute) and blood pressure (BP) of 110/70 mmHg. The abdomen was globular and followed the ventilatory movements, with no change in air sounds, soft, not contracted, depreciable, painful to coughing, and superficial and deep palpation in the left hypochondrium and hypogastrium. There was also a palpation of a hardened, hardened area of tumor configuration, fixed to internal planes, with no changes in skin colouring and a clear peritoneal reaction. As for the nervous system, a painful facies and a state of manifest anxiety were observed. No other apparent alterations were recorded.



Complementary laboratory tests

Complementary laboratory tests were performed, which are shown together with their normal reference values (NRV): leukocytosis at $14.2 \times 10^9/l$ (NRV= $5-10 \times 10^9/l$), hematocrit indicating mild anemia 0.34/l (NRV= 0.36- 0.44/l). Minimal coagulogram and other values within the normal range: platelets at $300 \times 10^9/l$ (VR= $150-400 \times 10^9/l$) and fasting blood glucose at 5.3 mmol/l (VR= 3.9-5.6 mmol/l).

Imaging examinations

A plain chest X-ray with PA (posteroanterior) view showed no pleuropulmonary, mediastinal, cardiac or osteomyoarticular alterations.

Abdominal ultrasonography was performed and showed no lesions in solid viscera or free fluid in the abdominal cavity; visceral dimensions were normal.

Soft tissue ultrasound revealed two pseudotumoural images with echogenic borders and echolucent centre, not septated, measuring approximately 84x30 mm and 36x21 mm respectively; they covered the planes of the anterior abdominal wall up to the aponeurosis.

Computed Axial Tomography (CT) of the abdomen showed a tumor of 42x34 and 94x46 mm, respectively, in the abdominal wall with an average density of 34UH, which appeared to belong to the muscular plane.

Due to the findings mentioned above, it was decided to schedule surgery for excision and biopsy. With the patient in the supine decubitus position, a Tilet was performed and the surgical site was prepared. The patient was started with a laparoscopic incision following Langer's lines, with subsequent excision of the tumor lesion (Figures 1A and 1B), leaving an oncological margin and exhaustive hemostasis. The wound was closed in planes and the wound was drained for follow-up.



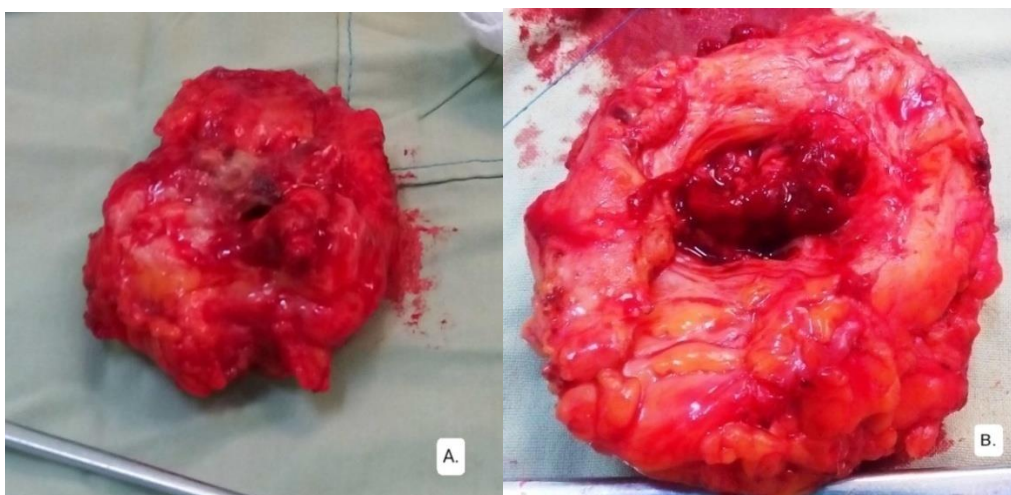


Fig. 1A and 1B. Pseudotumoral lesions of the anterior abdominal wall A. 42x34 mm. B. 96x46 mm. Note in both the central area of necrosis and the firm borders

He was transferred to the general surgery ward where he underwent the postoperative process without difficulty or local or general complications.

He received antibiotic treatment with ceftriaxone (1g) at the rate of two intravenous (IV) bulbs diluted in 20 cc of 0.9% physiological saline solution (PSS) every 12 hours. In addition, amikacin (500 mg) one intravenous bulb diluted in 200 ml of 0.9% SSF per day for one hour and metronidazole (500 mg), one EV bottle every 8 hours for 30 minutes each. It was decided to discharge the patient 32 hours postoperatively and to follow up with outpatient care in the Primary Health Care area.

The pathological anatomy service reported the formation of fatty tissue measuring 7x5.5 cm on section. A cystic lesion with sphacelated walls and a dirty semi-liquid content was observed in the thickness of the tissue. Another tissue formation measuring 8x5x3 cm, also consisting of fatty tissue was observed in the thickness of which a hemorrhagic and friable area was observed. No tumours were found in either of the two samples.

The subacute inflammatory cytological smear was negative for neoplastic cells.

Both slides, stained with haematoxylin and eosin, corresponded to severe chronic panniculitis with areas of granulation tissue where *Actinomyces* colonies were observed (Figures 2A and 2B).



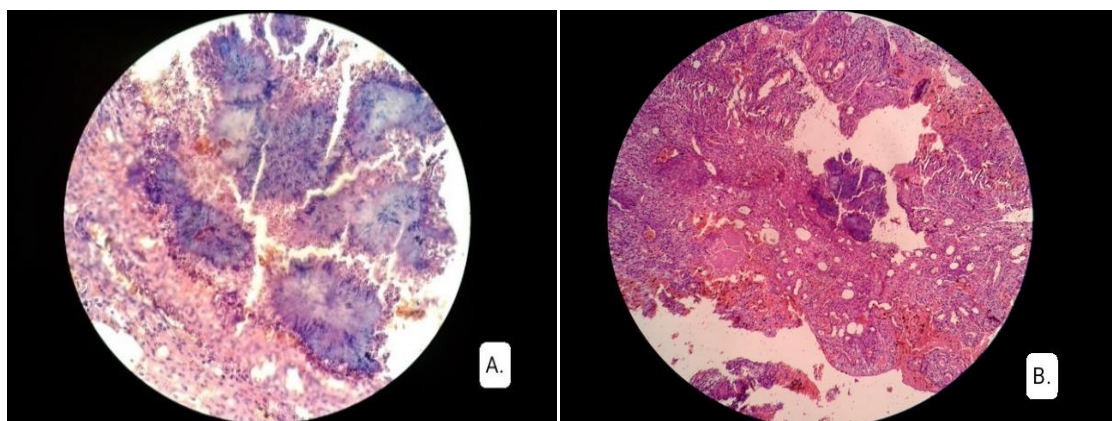


Fig. 2A and 2B. Histological images showing different views of areas of severe chronic panniculitis with granulation and Actinomyces colonies.

CASE DISCUSSION

Actinomyces bacilli are colonizers of the oral cavity, gastrointestinal tract and female genital tract. Cervicofacial actinomycosis is the most common clinical form; however, these infectious agents can affect any organ of the body and even the osteomyoarticular system.⁽⁷⁾ The abdominal location is uncommon, which makes the case presented here different from that described in published sources and of greater relevance.

In terms of diagnosis, CT or MRI may describe the presence of a thick-walled, infiltrative, irregular mass with one or more central hypodense areas that may be associated with fistulas, erosions or bone destruction. Differential diagnoses should therefore include neoplasms.⁽⁸⁾ However, these examinations only reported the presence of one tumor area in the patient and soft tissue ultrasound was able to identify areas of variable reflectivity and density compatible with pseudotumours, which may be explained by the location of the lesions.

Isolation of Actinomyces by culture occurs in 30-50 % of cases, because they are demanding bacteria that require media enriched with 6 % or 10 % CO₂.⁽⁷⁾ In the present case, isolation was not possible however, it is suggested that these agents can be observed in histological samples by haematoxylin-eosin staining.^(7,9) An element that corresponds to the case presented as it provided the definitive diagnosis.

Prolonged antibiotic treatment with several antimicrobials is the most commonly used, according to expert opinion and the results in several patients.⁽¹⁰⁾ In a case by Corrales-Valenzuela et al.⁽¹¹⁾ treatment focused on amoxicillin with clavulanic acid 500/125 mg every eight hours orally for seven months. The combination of intravenous penicillin G sodium and oral amoxicillin for six months to one year was also proposed as an effective treatment.⁽¹²⁾ Although these were not the combinations used, there was evidence of effectiveness in combining antibiotics and achieving clinical improvement.



As a possible risk factor triggering the infection; the presence of an IUD for more than seven years may have caused the growth and spread of the colonies and explains the unusual form of presentation in the abdominal wall due to its proximity to the gynecological system. There are documented cases where actinomycosis behaved in the same way, with the mucosa of the female genital system being one of the most frequent sites.⁽¹³⁾

FINAL CONSIDERATIONS

Actinomycosis is a bacterial infection caused by various genera of the *Actinomyces* family. The localization of colonies in the abdominal wall is uncommon and the diagnosis of certainty is made by obtaining a surgical biopsy and observation on histological slides.

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