

Cutaneous Botryomycosis Due to a Donkey Bite: A New Case

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Article History

Received: 04.12.2018

Accepted: 13.12.2018

Published: 30.12.2018

DOI:

10.36347/sjmcr.2018.v06i12.015



Abstract: Botryomycosis is a chronic granulomatous bacterial infectious suppurated pathology. It is rare and can be localized on the skin and the viscera. The predisposing factor is most often a trauma. This case is about a 28 year-old patient with no particular pathological history, who was bitten by a donkey at the age of 7, on the anterior side of the right leg. For the last 10 years, she had a right lower limb swelling, made of an atypical prurigo nodular-like, suppurative lesion with fistulas. There is also a functional impotence of the limb. The mycological samples returned sterile. Bacteriological analysis of cutaneous biopsies showed *Staphylococcus aureus*. Histological examination revealed granulomatous grain abscess infection, suggestive of more bacterial grains, in favor of a botryomycosis. The patient was treated with bi-antibiotic therapy adapted to the sensitivity of *S. aureus*, including ciprofloxacin and sulfamethoxazole / trimethoprim. We also added weekly applications of trichloroacetic acid at 50% at the vegetative fistula lesions, with good results after 4 months of treatment. Cutaneous botryomycosis is a rare and underrated unit. Our case is particular because the donkey bite has not yet been described as being responsible for botryomycosis. Trauma such as a snake bite, intravenous long-term infusion, and a fork injury were reported. Making diagnosis may be difficult as it requires clinical, biological and especially histological features. Differential diagnosis must be eliminated, especially fungal mycetoma and actinomycosis. *Staphylococcus aureus* is the most incriminated, but there are often several germs at the same time (aerobic / anaerobic) which makes it difficult to sterilize the lesions. Histopathologic findings, like an inflammatory infiltrate made of granuloma with basophilic center of granular mass and eosinophilic periphery (called: Splendore-Hoeppli phenomenon, non-pathognomonic), with a clinical history may help for the diagnosis. Finally, antibiotic therapy might help sometimes to sterilize lesions but in this case, trichloroacetic acid seemed effective in drying lesions.

Keywords: Cutaneous botryomycosis, donkey bite, bacterial infectious pathology, *Staphylococcus aureus*, Splendore-Hoeppli phenomenon, chronic granulomatous disease.

INTRODUCTION

Botryomycosis is a chronic, suppurated granulomatous pathology, due to several genera of bacteria [1]. Before, it was called by other names: staphylococcic actinophytosis, granular bacteriosis, actinobacillosis, and bacterial pseudomycosis [1]. This disease is rare and can be localized on the skin and internal organs. Its occurrence is favored by predisposing factors, most often a skin trauma. We describe a new case of a patient bitten by a donkey at the age of 7, in the right leg and having presented for 10 years, a cutaneous botryomycosis on the same lower limb.

CASE REPORT

A 28-year-old woman, at the age of 7, was bitten by a donkey on the anterior side of the right leg. She did not present with any other particular pathological history. For ten years, she has been developing a swollen right limb, with an atypical aspect of nodular prurigo-like lesions (ranging from the lower 1/3 of the leg to the lower 1/3 of the thigh), with fistulas budding lesions in places (Fig-1). The patient maintained a good condition without fever. In addition, there was a depressed scar on the anterior side of the leg (sequel of the bite) and an analgesic knee flexion and functional impairment of the limb.



Fig-1: right leg volume increased, aspect of nodular prurigo-like with fistulas budding lesions in places

HIV serologies, and hepatitis C and B were negative. There was no diabetes. A biological assessment showed hypochromic microcytic anemia (hemoglobin level at 9 g/dl) and iron deficiency. The search for primary immunodeficiency was negative. Bacteriological analysis of cutaneous biopsies showed *Staphylococcus aureus* and the search for *Actinomyces* germs was negative. Ziehl-Neelsen stain, research bacillus Koch, and fungal culture were negative too. Histopathological examination of a cutaneous biopsy showed a hyperplastic spongiotic epidermis, a dermis formed by a loose granulation tissue rich in congestive

vessels and including numerous siderophages, neutrophils, forming abscesses in places, histiocytes, lymphocytes, some eosinophilic polynucleus. With the presence of epithelioid granulomas with abscessed centers, small eosinophilic grains at the periphery, at the basophilic center with a microbacillary appearance. Complements of staining showed the presence of gram-positive cocci. This analysis concluded that a granulomatous grain abscess infection which is more reminiscent of bacterial grains is “in favor” of a botryomycosis (Fig-2).

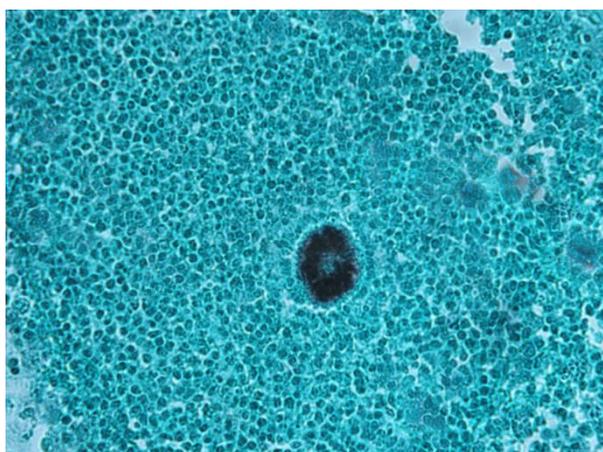


Fig-2: epithelioid granulomas with abscessed centers, small eosinophilic grains at the periphery, at the basophilic center with a microbacillary appearance

Standard radiography did not show any specific bone abnormalities or any appearance of intralesional foreign bodies. The patient was treated with a bi-antibiotic adapted to the sensitivity of *S. aureus*, ciprofloxacin and sulfamethoxazole / trimethoprim and a weekly applications of

trichloroacetic acid at 50% in the fistulas budding lesions.

During the 4 months of treatment, with regular monitoring of liver and renal functions, the patient has recovered well: the lesions dried up and the limb's volume decreased (Fig-3).

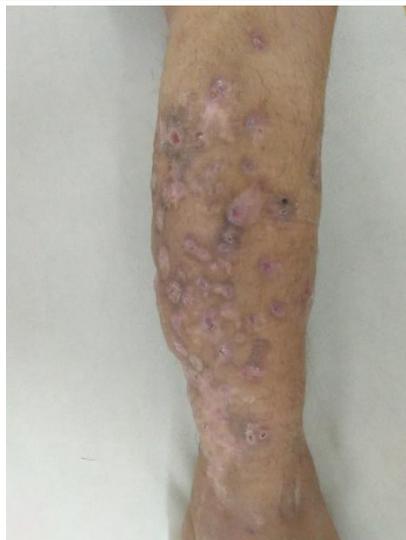


Fig-3: Drying of lesions and decrease in leg volume after 4 months of treatment

DISCUSSION

Botryomycosis is a rare and underrated entity [2]. It has been reported in several countries around the world, but its incidence and prevalence are poorly defined. Currently, about 200 cases have been documented, and cutaneous form represents approximately 75% of those reported cases [2, 3]. The cutaneous botryomycosis's physiopathology remains little known. It is due to an imbalance between the immune state of the host (a failure of probable cellular immunity and favoring factors) and the inoculation of the germ (importance of the bacterial load and the presence of a wound) [3, 4].

A previous wound is often incriminated as a precursor of the infection. A wide variety of traumas such as animal bites, including snakes, intravenous long-term infusion, and injury by a hay fork have been reported [1]. Chronic illness can also increase the risk of infection: diabetes mellitus, a retroviral infection, liver failure, a long-term corticosteroid, alcoholism, malnutrition or cystic fibrosis [3]. In our case, the skin trauma was the bite of a donkey, as being responsible for cutaneous botryomycosis. A bite of a donkey or an animal of the equine family can cause polymicrobial wounds with aerobic and anaerobic organisms, including the family of *Actinobacillus* and *Staphylococcus* (*hyicus* and *aureus*) [5, 6]. Studies have shown that these germs are found in the commensal flora of these equidae's nasal and oral secretions, which are non-pathogenic to the animal but may be the source of infection with other species, in particular the human [5]. The low initial bacterial inoculum is potentially the cause of the long incubation period in our case especially as the patient had no underlying illness.

Clinically, cutaneous botryomycosis mainly affects the limbs. It can be single or multiple, in the form of fistulized nodules most often. There are sometimes ulcerations, vegetative, verrucous aspects, or

prurigo-like. Bone and muscle extensions are possible. It usually evolves chronically, in a context of apyrexia, sometimes with pain or pruritus.

The diagnosis is often difficult: it requires clinical, biological, and especially histological arguments. Differential diagnosis are fungal mycetoma, nocardiosis and actinomycosis. When it is localized in the viscera, it can reach the brain, the kidney, the spleen and also the liver [4].

Botryomycosis belongs to a group of diseases that produces grains, which includes eumycetoma, actinomycetoma, actinomycosis, and botryomycosis. The difference between them is that eumycetoma grains are fungi. Actinomycetomas are aerobic actinomycetes that are filamentous, gram-positive, and partially acid alcohol resistant (AAR). Actinomycosis grains are anaerobic gram-positive actinomycetes. And finally, botryomycosis grains are true non-filamentous aerobic, anaerobic, gram-positive, and gram-negative bacteria [1, 3].

In 40% of cases [1], *Staphylococcus aureus* is the most common germ, but a polymicrobial flora is often found (aerobic / anaerobic), which makes it difficult to sterilize lesions [7]. The anatomopathologist must be experienced and know how to distinguish it from an inflammatory infiltrate made of granuloma with basophilic center of granules and eosinophilic periphery (phenomenon of Splendore-Hoeppli, which is non-pathognomonic).

Therapeutically, there is no consensus. Antibiotic therapy helps to sterilize lesions, and must be adapted to isolated germs and antibiogram. Surgery and drainage may be helpful, depending on the extent of the lesions. In our patient, the lesion remained cutaneous, despite the analgesic flexion of the knee and functional impotence. In addition to the antibiotics, the application

of trichloroacetic acid (50% concentrated) seems effective in drying lesions. The correction of underlying defects or contributing factors will be part of the management of the pathology.

Our patient progressed very well after a 4-month period of treatment with sulfamethoxazole / trimethoprim and ciprofloxacin, with regular monitoring of her renal and hepatic function.

CONCLUSION

The cutaneous botryomycosis is rare and unknown, to establish a correct diagnosis it is necessary for an early and adapted care. The history of skin injury must be sought. A clinical aspect of nodules with fistulas is often the presenting symptom. Bacteriological and fungal analyzes of the cutaneous samples must be thorough and oriented, to eliminate differential diagnosis. Histology is also helpful. Adequate antibiotic treatment should be initiated as soon as possible to prevent extension of the lesions and functional impotence.

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