Case Report

DOI: https://dx.doi.org/10.18203/2320-6012.ijrms20232161

Is skin grafting in the patient with cutaneous sporotrichosis the definitive therapy?

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Received: 18 June 2023 Accepted: 03 July 2023

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ABSTRACT

Sporotrichosis is a subacute to chronic infection caused by the dimorphic fungal genus Sporothrix. The infection usually affects the skin and subcutaneous tissues, but occasionally it can occur at other sites, mainly in immunocompromised patients. The symptoms of extracutaneous sporotrichosis can be subtle and diagnosis is often delayed. A 54-year-old male was received, originally from Huajuapan de León, Oaxaca; who was with an established diagnosis of type 2 diabetes mellitus; later admitted by the general surgery service with a diagnosis of necrotizing fasciitis of the left and right thoracic limb. When culture was collected with Sporothrix schenckii report, management was established with itraconazole, potassium iodide and with subsequent addition of amphotericin B, with antibiotic therapy directed for superinfection by opportunistic agents. After multiple surgical cleanings with degradation of necrotic tissue, implementation of negative pressure system and amputation of nonviable fingers, it was decided to proceed with the application of skin autografts, despite the persistence of the agent in subsequent culture reports, which are fully integrated, without evidence of new ulcerodular lesions so far. Although there are currently no parameters defined in the literature that guide the time or the appropriate conditions to perform skin grafts in the areas affected by cutaneous sporotrichosis, there are, on the contrary, reports of successful cases where surgical management has been effective even in the presence of positive cultures for Sporothrix schenckii. We consider that these results open the panorama of options for the current management of cutaneous sporotrichosis, making it necessary to consider surgical management in the therapeutic range of the same, allowing the deliberation on other more conservative options before the failure of classical therapy and the imminence of amputation.

Keywords: Iimmunocompromised, Surgery, Leukocytosis, Sporothrix, Infection

INTRODUCTION

Sporotrichosis is a subacute to chronic infection caused by the dimorphic fungal genus *Sporothrix*. The most common cause of infection is *Sporothrix schenckii*. The infection usually affects the skin and subcutaneous tissues, but occasionally it can occur at other sites, mainly in immunocompromised patients.¹ Subcutaneous mycoses refer to infections caused by fungi found in nature, acquired by direct inoculation and limited to the dermis and subcutaneous cellular tissue, but can spread to the epidermis or deeper planes, compromising bone structures, as well as spreading and producing systemic disease.² Sporotrichosis usually develops as a cutaneous syndrome following inoculation of fungi from soil or other organic material.¹ Extracutaneous forms of sporotrichosis may occur in isolation or as a manifestation of more widespread disease. The symptoms of extracutaneous sporotrichosis can be subtle and diagnosis is often delayed.³

While the typical host is a healthy individual with an outdoor occupation or hobby (e.g., landscaping, gardening) that provides exposure to the fungus; on the contrary, there are patients who, in addition to AIDS, frequently report other conditions such as diabetes, alcoholism, granulomatous diseases, cirrhosis, kidney transplantation, malignant neoplasms, use of corticosteroids and use of immunosuppressive agents; in any case, patients with HIV and preserved immunity appear to respond to infection in the same way as individuals without coinfection.^{4,5} Days or weeks after cutaneous inoculation of the fungus, a papule develops at the site of inoculation. This primary lesion is usually ulcerated, but may remain nodular with overlying erythema; The drainage of the lesion is not very purulent and has no odor. Similar lesions subsequently occur along the lymphatic channels proximal to the original lesion, a finding called sporotricoid dissemination or nodular lymphangitis. The nodules eventually rupture, ulcerate and discharge pus. Primary skin lesions may be mistaken for neoplasm and surgically а removed. Microscopically, these lesions are characterized by hyperkeratosis, parakeratosis and various degrees of pseudo-epitheliomatous hyperplasia and ulceration of the epidermis.⁴ The pain is generally mild and systemic symptoms are usually absent.⁵ Symptoms do not resolve without treatment.

CASE REPORT

In July 2022, a 54-year-old male from Huajuapan de León, Oaxaca, was received in a third-level hospital in the private sector of the city of Puebla; who had an established diagnosis of type 2 diabetes mellitus, during whose stay infection with the human immunodeficiency virus (HIV) was ruled out; subsequently admitted and consulted by the general surgery service with a diagnosis of necrotizing fasciitis of the limb. left and right thoracic.

He began his condition 4 months prior to his admission with presence of edema, and deformity of thoracic extremities, in end presence of interdigital flattens and on back of hand was observed; same that evolved to ulcers with fetid purulent exudate in abundant quantity, so went to doctor who established oral antibiotic management not specified without observing improvement, so in July 2022 he sought assistance from a dermatologist who decided to proceed with biopsies of right arm lesions, obtaining histo-pathological diagnosis of necrotizing fasciitis and *Phialemonium* sp infection, so he referred to hospital center for comprehensive management.

This is how he enters through the emergency department of our unit, from where he inter-consults the general surgery service, establishing management for necrotizing fasciitis by performing surgical toilets and taking subsequent cultures; during his stay he is also assessed by the plastic and reconstructive surgery service due to the significant structural damage to the tendon apparatus of the left hand, establishing the in viability.

Thirteen days after admission, surgical cleaning was performed with amputation of the fourth and fifth fingers

of the left hand with preservation of the base of muscle insertion of the rest of the phalanges, as well as surgical cleaning of the right hand, with placement of a negative system in both extremities; observing pressure predominance of abundant necrotic tissue in extensor tendons, scarce fibrin cream, exposure of tendons of second and third fingers of right hand and indications of granulation tissue in them, as well as presence of multiple ulcerodular lesions, characteristics of Sporothrix schenkii infection. Negative pressure system replacement was performed in both thoracic extremities and cultures were taken from the ulcerodular wounds described.

Three days later, a final report of wound culture was obtained reporting the presence of Klebsiella pneumoniae, Escherichia coli. Pseudomonas aeruginosa and highlighting the abundant growth of Sporothrix schenckii, this being the first report obtained from this agent. Subsequently, management was established with meropenem at 1 gram every 8 hours, levofloxacin 750 milligrams every 24 hours and antifungal treatment based on Itraconazole 200 milligrams every 24 hours.

Surgical cleanings were continued weekly, in addition to removal and placement of negative pressure system, as well as antibiotic therapy with itraconazole until day 39 and vancomycin on day 21 of effective administration, continuing antifungal therapy directed against S. schenckii with saturated potassium iodide solution orally at initial doses of 1 gram / day, climbing to reach 3 grams/day. Two months after admission, the infectology service reported the presence of Phialemonium sp. in culture of abscess of the right extremity, so it was indicated to start antibiotic therapy with amphotericin B calculated at 3 ml/kg/day. Three days after this report, a new surgical cleaning was performed, proceeding to the removal of the negative pressure system without complications and collecting cultures of persistent ulcerodular lesions.

After seven days of removal of the negative pressure system of both thoracic limbs, results of last cultures were collected, reporting, together with the persistence of S. schenckii, presence of Klebsiella pneumoniae in the right hand, sensitive to amikacin; and in the left-hand presence of Staphylococcus epidermidis sensitive to linezolid. tetracycline and vancomycin; therefore, antibiotic therapy with amikacin and vancomycin was initiated. Likewise, an increase in serum creatinine was evidenced in control laboratories, suggesting acute kidney injury KDIGO III and the necessary adjustments were made in the dosage of medication previously established based on potassium iodide and amphotericia B. Given the stabilization with these adjustments, it was not necessary to proceed with renal replacement therapy.

Five months after his admission, when the control radiography of thoracic limbs showed data of osteolysis at the level of the third finger of the left hand, the same

was amputated, raising the urgent need to assess additional therapeutic options to the classic management of cutaneous sporotrichosis by antifungals and debridement of nonviable tissue. This is how management is offered by taking autologous skin grafts, despite the persistence of presence of the agent in collected cultures, given the knowledge of success reports in similar cases in clinically clean margins. Thus, in November 2022, after five months of poor response to pharmacological management against sporotricosis, after assessing viability of underlying musculotendinous system, we proceed to perform autologous meshed skin graft of total thickness in both thoracic limbs, with taking them from the anterolateral face of ipsilateral pelvic limbs, deciding at same time the suspension of all antimicrobial/ antifungal therapy previously administered, continuing only with administration of oral antidiabetic drugs for control of its underlying pathology.

Fourteen days after the application of grafts, once their integration and adequate evolution have been verified, it is decided to discharge them from hospital with follow-up by outpatient consultation, obtaining adequate response to management, with total integration of the same and receiving discharge by general surgery service of hospital after eight months from the knowledge of their case.



Figure 1: Left hand wrist grafts.



Figure 2: Grafts in the dorsal region of the right hand.



Figure 3: Evolution of the grafts in both hands 1 month after the surgeries.

DISCUSSION

In general, when sporotrichosis is suspected, a biopsy culture of tissue, sputum, body fluids or aspirated material from lesion should а skin he usually collected. Histopathology performed is simultaneously.¹ Itraconazole has been used effectively and safely in most cases of sporotricosis, with low toxicity and good tolerance, even in long-term treatments, however, resistance of infection to it has also been documented in isolated cases.⁴

In a study conducted in Brazil following an epidemic of lymphocutaneous sporotrichosis between 2002-2006, of a total of 645 patients, six hundred and ten patients (94.6%) were cured with itraconazole (547 at doses of 100 mg/ day, 59 with 200-400 mg/ day and 4 children with 50 mg/ day). The median treatment time was 12 weeks with disseminated lymphocutaneous and cutaneous forms. Patients using antacids had a median time to longer cure (14.5 weeks), regardless of clinical form.⁶

CONCLUSION

Although there are currently no parameters defined in the literature that guide the time or the appropriate conditions to perform skin grafts in the areas affected by cutaneous sporotrichosis, there are, on the contrary, reports of successful cases where surgical management has been effective even in the presence of positive cultures for Sporothrix schenckii. In our case, a male patient of the sixth decade of life, with risk factors for sporotrichosis infection, peasant by trade, presents to a tertiary center after the histopathological diagnosis of necrotizing fasciitis in both upper extremities, being managed for such entity until the finding of infection by Sporothrix schenckii, from where the classic management for the agent based on potassium iodide and itraconazole begins, with poor response to it and important advance of the extension of ulceronodular lesions as well as deep tissues, reaching the bone tissue and making necessary the amputation of fingers of the left hand. When growth of Sporothrix schenckii was obtained in the culture media and given the relative failure of conventional therapy, after stabilization of the patient and based on the described reports of surgical management through skin grafts in clinically clean margins, it was decided to opt for this alternative by withdrawing the administration of all medication, resulting, finally, successful, even in the face of the persistence of positive crops.

We consider that these results open the panorama of options for the current management of cutaneous sporotrichosis, making it necessary to consider surgical management in the therapeutic range of the same, allowing the deliberation on other more conservative options before the failure of classical therapy and the imminence of ablation.

Funding: No funding sources Conflict of interest: None declared Ethical approval: Not required

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Cite this article as: Monterrubio NML, Sánchez OS, Montaño MH, Barrientos CZD, Tovar FN, Trujillo HAC. Is skin grafting in the patient with cutaneous sporotrichosis the definitive therapy? Int J Res Med Sci 2023;11:3054-7.