Lymphocutaneous Sporotrichosis of Face with Verrucous Lesions: A Case Report

Abstract

Sporotrichosis is a cutaneous mycosis caused by a dimorphic fungus, *Sporothrix schenckii* species complex clinically presenting as lymphocutaneous, fixed, or disseminated forms. A typical lesion is an erythematous papule, noduloulcerative lesion usually occurring at the site of penetrating trauma, mostly on the extremities. Verrucous lesion is an unusual presentation of sporotrichosis which can mimic the verrucous lesions seen in chromoblastomycosis, tuberculosis verruca cutis/lupus vulgaris (TBVC/LV), cutaneous leishmaniasis, and blastomycosis leading to diagnostic dilemma. Herein, we describe a case of facial verrucous sporotrichosis in a child from sub-Himalayan region.

Keywords: Itraconazole, lymphocutaneous, sporotrichosis, verrucous

Introduction

Sporotrichosis is a chronic granulomatous subcutaneous mycotic infection caused by dimorphic fungus *Sporothrix schenckii* species complex, a common saprophyte of soil, sphagnum moss, and plant detritus. The disease is endemic in regions with a tropical or subtropical climate and among personnel involved in agriculture or handling of plant material. After traumatic implantation of the pathogen into the skin, the disease develops as fixed cutaneous, lymphocutaneous, or disseminated cutaneous sporotrichosis mainly involving extremities in its classic forms. Lymphocutaneous sporotrichosis is the most common form accounting for a majority cases where diagnosis is fairly easy. Facial involvement occurs mostly in fixed cutaneous variety and among childhood cases. Clinically, the lesions are usually asymptomatic, erythematous, papules or papulo-pustules, nodules with or without ulceration, non-healing ulcers, or small abscesses. Rarely, lesions of verrucous morphology are seen, especially in long-standing cutaneous sporotrichosis. This poses a diagnostic dilemma particularly in nonendemic regions or where cutaneous tuberculosis is prevalent leading to a misdiagnosis as tuberculosis verruca cutis (TBVC) or hypertrophic lesions of lupus vulgaris. Verrucous lesions of cutaneous sporotrichosis are also difficult to differentiate clinically from those of chromoblastomycosis, blastomycosis, cutaneous leishmaniasis, or viral warts. We report an uncommon case of facial lymphocutaneous sporotrichosis with verrucous lesions where clinical suspicion together with microbiological investigations helped in confirming the diagnosis and appropriate management.

Case Report

A 14-year-old boy from a rural background presented with a large warty lesion with minimal purulent discharge on the tip of the nose and ala nasi, as well as erythematous small plaques on the left cheek for 3 months. The lesion on the tip of the nose started as small asymptomatic erythematous papule which gradually increased to attain the present size, and later became ulcerated, crusted and warty, and enlarged to involve the adjoining left nare and ala nasi [Figure 1]. He also developed two more nontender, well-defined, erythematous, papulo-plaques over the left cheek along the lymphatics [Figure 2]. He denied any history of insect bite, local trauma, or traumatic manipulation of the initial lesion, or staying in areas endemic for cutaneous leishmaniasis. His medical and family history were unremarkable. He had received repeated treatments with antibiotics at a health center without improvement.
improvement. Hair and nails were normal. General physical and systemic examination did not reveal any abnormality. He was further investigated with the clinical diagnosis of chromoblastomycosis, TBVC, lupus vulgaris, and cutaneous leishmaniasis. Routine biochemistry and hematological investigations including chest X-ray were normal. Mantoux test was negative. Repeated examination for yeast cells and/or sclerotic bodies in 10% KOH mounts from lesional scrapings or Leishman Donovan (LD) bodies in Giemsa-stained tissue smears yielded no organism. A skin biopsy from the lesion showed pseudoepitheliomatous hyperplasia with micro-abscesses formation and chronic granulomatous inflammatory infiltrate comprising lymphocytes, histiocytes, epithelioid histiocytes, and occasional multinucleated giant cells [Figures 3 and 4]. Biopsy specimens subjected to culture on LJ medium grew no pathogen. Growth of characteristic white to grayish colonies were seen at 1 week after incubation at 25°C on Sabouraud’s dextrose agar (SDA) containing chloramphenicol and cyclohexamide. These colonies became grayish black on prolonged incubation [Figure 5]. *S. schenckii* species complex was identified from colony characteristics by temperature dimorphism and conversion to yeast form by subculture on brain heart infusion blood agar and incubation at 37°C for 7–10 days [Figure 6]. Characteristic thin hyaline septate hyphae, sessile pigmented conidia with rosette arrangement in lactophenol cotton blue (LCB) mounts were noted [Figure 7]. The isolate could not be confirmed by gold standard molecular

![Figure 1: The verrucous plaque with ulceration and black crust, extending to the left side of the nose tip, ala nasi, and anterior nasal septum sparing nasal mucosa. Note two erythematous papulo-plaque lesions over the left cheek in a lymphocutaneous pattern](image1)

![Figure 2: Another view of verrucous plaque over the nose tip and left ala nasi. Note other two lesions over the left cheek](image2)

![Figure 3: Hematoxilin and eosin staining of tissue showing pseudoepitheliomatous hyperplasia with micro-abscess formation (magnification ×400)](image3)

![Figure 4: Hematoxilin and eosin staining of tissue showing micro-abscesses surrounded by lymphocytes, histiocytes, and occasional multinucleated giant cells (magnification ×1000)](image4)
techniques due to unavailability. This is a limitation of the present case report. The lesions healed completely after treatment with itraconazole 100mg two times a day and supersaturated solution of potassium iodide (SSKI) 5 drops three times a day which was increased to 20 drops three times a day for 16 weeks without significant adverse effects [Figure 8].

Discussion

A painless papule develops at the site of traumatic inoculation of the fungus, which progresses to a nodular lesion both in lymphocutaneous and fixed cutaneous sporotrichosis. This usually evolves into a nodule, or noduloulcerative, or erythematous plaque localized at the inoculation site in fixed cutaneous sporotrichosis, or new lesions develop along the lymphatics of lymphocutaneous variant. Our patient had classic presentation of lymphocutaneous sporotrichosis with primary lesion over the nose tip and new lesions developing over the left cheek along the lymphatics. The lesion may also

Figure 5: Sabouraud’s dextrose agar showing brown to black velvety, waxy colonies of Sporothrix schenckii

Figure 6: Gram-stained smear showing cigar-shaped yeast cells seen on mycelium conversion of S. schenckii after inoculation on BHI blood agar and incubation at 37°C (magnification ×1000)

Figure 7: Characteristic thin hyaline septate hyphae with pigmented sessile conidia of S. schenckii in rosettes arrangement (Lactophenol cotton blue stain, ×40)

Figure 8: Completely healed lesions with post inflammatory hyperpigmentation over the nose and cheek after 16 weeks of treatment
develop into a verrucous nodule enlarging to a warty plaque often leading to diagnostic confusion of chromoblastomycosis or TBVC. The demonstration of sclerotic bodies on KOH mounts and culture of causative fungus in SDA is essential for the diagnosis in chromoblastomycosis cases. However, TBVC/LV, a paucibacillary *Mycobacterium tuberculosis* infection of the skin, being common in India was distinctly possible in this case, but growth of *S. schenckii* on SDA was diagnostic.[6]

Verrucous lesions in chronic cutaneous sporotrichosis have been reported uncommonly. A 16-month-old Korean girl had verrucous plaque on the dorsum of the hand while another 7-year-old boy developed a verrucous sporotrichosis over the chin after a bicycle accident.[3,4] Both the patients were treated successfully with itraconazole given for 14 weeks. Carr et al. also reported a case of extensive verrucous sporotrichosis involving the entire lower extremity mimicking cutaneous blastomycosis.[2] Motswaledi et al. described another case of a diabetic African man with a warty lesion on the back and treated with oral itraconazole 200mg two times a day.[7] It is interesting to know the pathogenesis of verrucous lesions in sporotrichosis and the reasons for its rarity. Pseudoepithelomatous hyperplasia due to some immune-mediated inflammatory reaction remains a possibility. Nevertheless, itraconazole 100–200 mg given twice a day is the treatment of choice for all forms of sporotrichosis, especially in the developed world, whereas SSKI has been used with consistent therapeutic response in resource-poor countries where majority of the cases occur.[8,9] However, SSKI acting synergistically with itraconazole in combination improves therapeutic outcome. Treatment with itraconazole in combination with SSKI given for 16 weeks achieved cure in our patient.

Although uncommon, verrucous lesions may occur in cutaneous sporotrichosis as well. These need to be differentiated from other common infective disorders (vide supra). High index of clinical suspicion, particularly in nonendemic regions, is imperative for accurate treatment and to prevent morbidity associated with delayed diagnosis.

**Contributions**

Author No. 1 has done laboratory work and written the article. Author No. 2 has treated the patient and written the clinical aspects of the article and author No. 3 has seen the patient, collected sample and edited the article.

**Declaration of patient consent**

The authors certify that they have obtained all appropriate patient consent forms. In the form the patient(s) has/have given his/her/their consent for his/her/their images and other clinical information to be reported in the journal. The patients understand that their names and initials will not be published and due efforts will be made to conceal their identity, but anonymity cannot be guaranteed.

**Financial support and sponsorship**

Nil.

**Conflicts of interest**

There are no conflicts of interest.

**References**

1. Chakrabarti A, Bonifaz A, Gutierrez-Galhardo MC, Mochizuki T, Li S. Global epidemiology of sporotrichosis. Med Mycol 2015;53:3-14.
2. Carr RD, Storkan MA, Wilson JW. Extensive verrucous sporotrichosis of long duration. Report of a case resembling cutaneous blastomycosis. Arch Dermatol 1964;89:124-30.
3. Kwon KS, Yim CS, Jang HS, Chung TA, Oh CK. Verrucous sporotrichosis in an infant treated with itraconazole. J Am Acad Dermatol 1998;38:112-4.
4. Williams BA, Jennings TA, Rushing EC, Wirges M, Russell BE. Sporotrichosis on the face of a 7-year-old boy following a bicycle accident. Pediatr Dermatol 2013;30:246-7.
5. James WD, Berger T, Elston DM, editors. Andrews’ Diseases of the Skin Clinical Dermatology. 10th ed. Philadelphia, USA: Saunders Elsevier; 2006.
6. Kwon-Chung KJ, Bennett JE. Sporotrichosis.In: Kwon-Chung KJ, Bennett JE, editors. Medical Mycology. 2nd ed. Philadelphia, USA: Lea and Febiger; 1992.p. 707-29.
7. Motswaledi H, Nkosi L, Mooba C, Ngobeni K, Nemutavhanani D, Maloba B. Sporotrichosis: A case report and literature review. J Clin Exp Dermatol Res [serial on the internet]. 2011 [cited 2011 Nov 25];2:[about 4 p]. Available from: https://www.pdfs.semanticscholar.org.
8. Ramirez Soto MC. Sporotrichosis: The story of an endemic region in Peru over 28 years (1985-2012). PLoS One2015;10:e0127924.
9. Pappas PG, Tellez I, Deep AE, Nolasco D, Holgado W, Bustamente B. Sporotrichosis in Peru: Description of an area of hyperendemicity. Clin Infect Dis 2000;30:65-70.