

SPOROTRICHOSIS IN CHILDREN: A FORGOTTEN ENTITY

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Abstract

Sporotrichosis is regarded as the most common subcutaneous fungal mycosis, which occurs following traumatic inoculation of *Sporothrix schenckii* complex. Albeit rare, sporotrichosis, also known as 'rose gardener's disease', has been reported to involve children whereby active or passive transmission through a vector such as domestic animals have been identified as the main culprit. Herein, we report sporotrichosis in a pair of siblings who presented to the outpatient Otorhinolaryngology clinic in University Malaya Medical Centre with a lower lips skin lesion and submental swelling after kissing their domestic cat. Sporotrichosis was diagnosed following the fungal culture of the skin scrapings of the lower lip's pustular lesions. Prompt treatment with oral itraconazole (8 mg/kg/day) for six weeks showed the resolution of symptoms. We highlight the pathophysiology of zoonotic sporotrichosis and the mode of management of this entity. Additionally, the management of sporotrichosis should include the veterinarian's assessment of the infected animal.

Keywords: Sporotrichosis, Sporothrix, Zoonosis, Skin lesions, Children, Lymphadenopathy

Introduction

Sporotrichosis, also known as 'rose gardener's disease', is a mycotic infection caused by the dimorphic fungus, *Sporothrix schenckii*. Albeit a worldwide distribution, it is considered endemic in temperate areas with high humidity (80–95%) (1). Sporotrichosis is regarded to be the most common subcutaneous mycosis. More than 6 species have been identified, including *Sporothrix schenckii sensu stricto*, *Sporothrix brasiliensis*, *Sporothrix globosa*, *Sporothrix mexicana*, and *Sporothrix albicans* were identified by molecular techniques. DNA topoisomerase II gene and M13 PCR fingerprinting of *Sporothrix Schenckii* corroborated its heterogeneity (1). Akin to all mycotic pathogens, virulence factors reported in Sporotrichosis include combination factors including temperature tolerance, evasion of hosts defence, dimorphism and enzymatic activities.

Nonetheless, the adhesion of the fungus to the host extracellular matrix has been advocated as the critical factor in the development of sporotrichosis (2). The rising number of human cases results from the increased number of cats. Moreover, high zoonotic transmission potential has been reported due to a high fungal load in the cat's skin lesion (3).

Sporotrichosis can possibly infect anyone regardless of gender and age, and it depends on the exposure (1). The One Health approach advocated by the World Health Organization: the collaboration between physicians

and veterinarians is indispensable to control zoonotic sporotrichosis outbreaks (4). We discuss our encounter with sporotrichosis in a pair of siblings.

Case report

Sibling 1

A 7-year-old boy presented to the Otorhinolaryngology Clinic at the University Malaya Medical Centre with a one-week history of papular lesions over the right lower lip. According to the mother, she noticed the skin lesion was extending to the chin. The rashes and gradual enlarging submental swelling were seen after 2 days. Over the past few days, the papular skin lesions transformed into pustular lesions, which ruptured after 1 week, producing a yellowish discharge. Subsequently, the submental swelling became painful and tender with no overlying skin changes, not warm to touch and non-fluctuant. However, there were no dysphagia, odynophagia, fever, or obstructive symptoms. No other rash was noted elsewhere in the body. The child was tolerating orally well with no vomiting or diarrhoea.

On examination, the child was comfortable under room air and not septic looking. Dried pustules were noted over the lower lips and chin (Figure 1). Intraoral examination was unremarkable. Neck examination revealed tender and firm submental lymph nodes measuring 1 x 2 cm. No other neck nodes were palpable. Lungs were clear on auscultation, and no hepatosplenomegaly was palpable.



Figure 1: Dried pustules over lower lip and chin

Sibling 2

Sibling 2 was a previously healthy 10-year-old girl with similar signs and symptoms. The papular lesions appeared to spread to the chin region with no discharge. Dried pustules were noted over the lower jaw after examining the second sibling. Intraoral examination was unremarkable. A neck examination revealed a tender, vague submental lymph nodes.

Both children were brought to a general practitioner clinic a week after their initial presentation and given antibiotics. However, no improvement was noted upon the completion of antibiotics.

Full blood count, liver function test, renal profile, erythrocyte sedimentation rate (ESR), and C-reactive protein (CRP) taken for both siblings were unremarkable. However, the ultrasonography neck for the brother revealed multiple enlarged lobulated lymph nodes seen at the submental region with fatty hilum collectively measuring 3.1 x 2.0 cm. As for the sister, neck ultrasonography revealed multiple enlarged lymph nodes, the largest measuring 0.7 x 1.4 cm. Scrapping of the lesion from the lower lip of both siblings performed revealed *Sporothrix schenckii*. Given the strong clinical history along with positive scrapping findings, a diagnosis of sporotrichosis was made, and neither fine needle aspiration cytology nor biopsy of the submental swelling was performed.

Differential diagnosis of multiple neck nodes with a history of cat exposure includes Cat-scratch disease as well as *Toxoplasmosis gondii*, which may present similarly clinically. However, histopathological examination and toxoplasma serology will differentiate each of these entities.

Both siblings were started on oral itraconazole (8 mg/kg/day) for 6 weeks following a discussion with the Paediatrics Infectious Disease team. The two-week outpatient follow-

up revealed both siblings to be responsive to the oral antifungal, and the submental swelling and the rashes reduced tremendously. The mother also brought the cat to the veterinarian. After 1 month, both siblings were seen again in the outpatient clinic, and the skin lesions were resolved entirely.

Discussion

Benjamin Schenk from Johns Hopkins Hospital was the first to describe sporotrichosis in 1898; subsequently, Hektoen and Perkins named the etiologic fungi *Sporothrix schenckii* in 1900 (5). The fungus was eventually named *Sporothrix schenckii*, and Guarro et al. (6) classified the fungus under the division Ascomycota, class Pyrenomycetes, order Ophistomatales, family Ophistomataceae.

Sporothrix schenckii can be found in the soil, decaying vegetation, plants, timber, hay, and moss. It is more commonly found in adults and can be occupational-related; for example, farmers are found to be at high risk for contracting the disease (1). Posttraumatic inoculation of the fungus has been reported to be the most common route of infection, although inhalation of spores is also possible despite being scarcely reported (7). Additionally, cases of sporotrichosis involving children have been less frequently reported as compared to adults (8). Case-controlled studies revealed playing in crop fields and on dirty floors is the main culprit of sporotrichosis among children (9).

In the same vein, animals can also act as vectors for disease transmission, notably cats, dogs, horses, camels, donkeys, or rats. The spread of disease from animals can be explained in two ways: either by direct transmission from a diseased animal, especially involving cats, or through passive transmission, whereby the disease is transmitted by an animal that is free from the disease. Passive transmission usually involves warm-blooded animals such as cats, dogs, birds, and parrots (9). Although the cats did not scratch both of our patients, we believe our patients contracted the disease via direct transmission from kissing the wound of the infected cat.

Based on the clinical aspect, there are cutaneous-lymphatic, fixed-cutaneous, disseminated-cutaneous, mucosal, and cutaneous hematogenous forms and extracutaneous forms, which include conjunctiva, pulmonary and osteoarticular. The cutaneous-lymphatic form of sporotrichosis, which commonly involves the facial region of the patient, is encountered in up to 40 % of all cases involving children, in which our patients also shared a similar presentation. In contrast, in adults, the disease mainly affects the extremities (9). The relatively thinner and delicate skin amongst children has been reported to have disseminated sporotrichosis in the facial region (6). Although an injured skin surface predisposes a child to Sporotrichosis transmission, transmission through intact skin has also been encountered (8), as reported in our

patients. It is too salutary to note that the incubation period of sporotrichosis varies from days to months (5).

A thorough history of occupation, travelling, hobbies, and animal contact is crucial in addition to high clinical suspicion to ensure prompt management. Although a battery of investigations is usually carried out prior to obtaining a diagnosis, it is noteworthy that direct examination of exudate, sputum, and synovial fluid is not helpful as the fungal structures are not usually observed (5). Histopathological examination can be helpful if fungal elements are observed, although the yeast forms of the fungus are rarely seen (5). Fungal culture has been regarded as the gold standard for the diagnosis of sporotrichosis (8). However, it is also important to note that a negative result does not exclude the possibility of sporotrichosis.

Sporotrichosis should be suspected if a localised, crusted papule, papuloulcerative or nodular lesion is persistent despite antibiotic administration (9). The differential diagnosis of sporotrichosis includes cutaneous tuberculosis, leishmaniasis, nocardiosis, chromoblastomycosis, blastomycosis, paracoccidioidomycosis, and non-tuberculosis mycobacteriosis (9). In our patients, no invasive diagnostic procedures were performed following positive results from the scrapping of the lower lip lesions. The children were clinically well and not in sepsis. Nevertheless, if the children did not respond to the treatment, a biopsy of the skin lesion and the submental swelling would be the next step.

As for the optimal mode of treatment, Potassium Iodide (KI) has been used to treat sporotrichosis since 1903 (10). KI is the drug of choice in many countries for lymphocutaneous and fixed sporotrichosis, given its efficacy, safety, ease of management, and low cost. However, itraconazole has been regarded as the first choice of treatment for sporotrichosis based on its effectiveness as well as safety profile (9). The dosage for itraconazole ranges between 100 and 200 mg per day, although, in relapsing or recalcitrant cases, the dosage can increase up to 300–400 mg per day. In children up to 20 kg of weight, itraconazole's dosage is 5–10 mg/kg/day (6). Both our patients were treated with itraconazole and showed improvement within two weeks of treatment. Albeit rare, intravenous Amphotericin B is the treatment of choice for systemic or disseminated cases, particularly when sporotrichosis involves the bone, visceral and pulmonary (9, 10). Thermoherapy, such as heat therapy, is useful when systemic antifungal agents are deemed unsuitable, especially amongst pregnancy-related disseminated cutaneous sporotrichosis (7).

Parallel to that, cats or any identified animals transmitting sporotrichosis should be assessed by the veterinarian and isolated until it is treated. Additionally, any physical contact with infected animals should be avoided until complete healing of the lesions is visualised (9).

It is also worth noting that organisations such as One Health, an integrated unifying approach that aims to balance and optimise the health of human, animals and

ecosystems, enables collaboration of multiple sectors and communities to manage entities such as sporotrichosis (4). The joint efforts between healthcare professionals such as veterinarians and physicians are crucial to ensure timely diagnosis, surveillance, and control of zoonotic sporotrichosis outbreaks.

Conclusion

Sporotrichosis, albeit common amongst adults, has been rarely reported amongst children. History of recent encounters, especially with domestic animals, is vital to obtain a diagnosis. Detailed clinical history and high suspicion of sporotrichosis are crucial for prompt diagnosis and management. Once the diagnosis is made and the child has been treated, the animal responsible or suspected to have transmitted, a veterinarian should evaluate sporotrichosis. Follow-up is important to watch for regression of the lesion.

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Competing interests

The authors declare that they have no competing interests.

Informed consent

It was obtained from the mother of the siblings.

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