## Clinical features of 10 cases of eyelid sporotrichosis in Jilin Province (Northeast China)

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#### ABSTRACT ● RÉSUMÉ

Purpose: Sporotrichosis is a common subcutaneous mycosis caused by an infection with dimorphic fungus Sporothrix schenckii. We present a series of patients with eyelid sporotrichosis and study the clinical and histopathological presentation, microbiology, treatment options, and outcome.

Methods: A retrospective case-series study of patients with a clinical diagnosis of eyelid sporotrichosis. Records were examined to obtain information regarding patient demographics, presenting symptoms and signs, histopathological examination, microbiology,

Results: Ten patients (4 men, 6 women; mean age 46.5 years, range 3-81 years) were included. Based on their clinical manifestations, eyelid sporotrichosis was classified into 3 major forms: (i) fixed cutaneous (6/10 cases), (ii) lymphocutaneous (3/10 cases), and (iii) eyelid abscess (1/10 cases). All the cases were treated with a terbinafine 12-week regimen. Nodules, papules, and abscesses regressed after treatment. No recurrence was discovered after a 12-week follow-up.

Conclusions: Eyelid sporotrichosis has typical features of clinical manifestations. Histopathological examination and tissue culture are helpful for diagnosis. Confirmed cases normally require long-term systematic treatment with antifungal agents, but surgical removal is normally unnecessary.

Sporotrichosis can be a subacute or chronic condition. This infection is caused by the dimorphic fungus Sporothrix schenckii, which has been isolated from straw, wheat grain, wood, and similar materials. Sporotrichosis is currently distributed throughout the world. In Canada, George Learmonth was the first to claim to have identified derma sporotrichosis infection and successfully cultured S. schenckii in 1911.2 In 1995, Carr et al. reviewed a series of 7 cases of sporotrichosis from various Toronto teaching hospitals and revealed that the medical history usually involved puncture injury contaminated with soil.3 In 1988, a large outbreak of sporotrichosis caused by handling conifer seedlings and involving 84 patients from 15 different states was also reported in the United States.<sup>4</sup> Since the end of the 1990s, there has been an epidemic of sporotrichosis associated with transmission by cats in Rio de Janeiro, Brazil. More than 2000 human cases and 3000 animal cases have been reported.<sup>5,6</sup>

In Jilin Province in northeastern China, people living in rural areas often heat their homes in winter by burning corn straw and wood. This is one of the major causes of the seasonal outbreaks of sporotrichosis that occur in this area. Among the increasing numbers of confirmed cases of infection, the eyelid has been found to be a very common site. Ophthalmologists are often the first medical practitioners to see patients with sporotrichosis. In this study, 10 confirmed cases were examined and used to summarize the clinical features and treatment of eyelid sporotrichosis.

#### **METHODS**

This study was performed according to the Declaration of Helsinki standards and was approved by the Research Ethics Committee. Written informed consent was obtained from all patients through mail or email. Medical reports of 10 patients who presented to the Second Hospital of Jilin University from April 2014 to April 2015 with a clinical diagnosis of eyelid sporotrichosis were retrospectively reviewed. All patients were from Jilin Province in northeastern China. The information that was recorded included patient demographics, presenting symptoms and signs, histopathological examination, microbiology, management, and outcomes. According to the clinical manifestations and previous studies of skin sporotrichosis, local eyelid sporotrichosis can be classified into the following 3 major forms: (i) fixed cutaneous, (ii) lymphocutaneous, and (iii) eyelid abscess.

#### RESULTS

Table 1 summarizes the demographics of eyelid sporotrichosis, presenting symptoms and signs, histopathologexamination, microbiology, management, and outcomes. Most patients were women (n = 6, 60%), and age ranged from 3 to 81 years (mean 46.5 years). Results collected in the present and previous studies suggest that eyelid sporotrichosis can be classified into 3 major forms based on their clinical manifestations: (i)

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			Se	ex			Histopathological Examination			
Clinical Type	Case Numbers	Age (years)	F	М	Presenting Time (months)	Presenting Signs	Granuloma- Positive Case Numbers	Asteroid Body- Positive Case Numbers	Tissue Culture– Positive Case Numbers	Treatment (weeks)
i	6	3–81	4	2	1–3	Nodular, ulcerative, plaque-like, or verrucous lesions 0.5–3 cm	6	4	5	TB 250 mg/d for 12 wk in combination with PI BID; for children, reduced dose
ï	3	15–62	2	1	1–2	Indurated papule progressively forms nodules, cysts, or ulcerations along the lymph trajectory	3	2	3	TB 250 mg/d for 12 wk in combination with PI BID
iii	1	44-53	0	1	1	Eyelid abscesses	1	1	1	TB 250 mg/d for 12 wk

fixed cutaneous, (ii) lymphocutaneous, and (iii) eyelid abscess. The fixed cutaneous form was found to be the most common form in the eyelid (6/10 cases). It is characterized by localized cutaneous lesions approximately 0.5–3 cm in diameter and 1 or 2 differently sized nodular, ulcerative, plaque-like, or verrucous lesions restricted to the site of inoculation (Fig. 1A–C). The lymphocutaneous form was also found to be common (3/10 cases). It usually presents an indurated papule that progressively forms nodules, cysts, or ulcerations, and further nodules appear along the lymph trajectory (Fig. 1D, E). In rare cases, S. schenckii infections can also produce eyelid abscesses (1/10 cases). These abscesses present as a tender mass with a diameter of about 1–3 cm (Fig. 1F).

Histopathological examination is helpful in suggesting the diagnosis. Characteristic sporotrichoid granulomas were present in most cases. These granulomas consist of a central microabscess with neutrophils and necrosis (Fig. 2A, B). Asteroid bodies, another histopathological characteristic change of *S. schenckii* infection, were observable in the centre of the granuloma in approximately 70% of cases (7/10 cases) (Fig. 2C, D). Moreover, the yeast cells were visualized directly with periodic acid-Schiff staining on tissue sections. The spore is an oval to cigar-shaped yeast, 3–9 µm in diameter, and the wall of each spore is double-layered. These cell walls stained purplish red (Fig. 3). A definitive diagnosis normally requires isolation of the fungus from tissue culture in Sabouraud agar. The organism grows readily on Sabouraud dextrose agar at 25°C, producing cream-colored colonies that mature into black, leathery colonies in a few days (Fig. 4).

All the patients were treated with the terbinafine 12-week regimen. Specifically, terbinafine 250 mg/day was prescribed to those whose body weight was over 40 kg. For those whose body weight was above 15 kg and less than 40 kg, the daily dose was 125 mg. For those whose body weight was less than 15 kg, 62.5 mg terbinafine was prescribed as the daily dose. Oral terbinafine may cause



Fig. 1—Clinical features of eyelid sporotrichosis. (A–C) Fixed eyelid sporotrichosis presents with papular, plaque-like, nodular, verrucous lesions that remain confined to the site of initial inoculation. (D, E) Lymphocutaneous eyelid sporotrichosis is characterized by the emergence of an indurated papule and further nodules appearing along the lymph trajectory contiguous to the initial lesion (black arrow). (F) Eyelid abscess is a tender mass with a diameter of about 1–3 cm; it is generally surrounded by an area of pink to deep-red discoloration.

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