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SPOROTHRIX SCHENCKII VAR LURIEI AS THE CAUSE OF SPOROTRICHOSIS IN ITALY

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The second known case of sporotrichosis caused by *Sporothrix schenckii* var. *luriei* in a patient living in Piacenza, Italy is described. In the absence of cultures, the diagnosis was based on histologic studies. Stained tissue sections (Hematoxylin and eosin, & Gomori methenamine silver) revealed hyaline, large, thick walled tissue form cells that had divided by septation or a budding process. These forms, along with the striking "eyeglass" configuration of incompletely separated cells that were also present, are the diagnostic features of this apparently rare variety. The use of a fluorescent antibody reagent, specific for *S. schenckii*, confirmed the identity of the etiologic agent.

INTRODUCTION

First described by Ajello and Kaplan in 1969 (1) Sporothrix schenckii var. luriei was the cause of a fixed type of sporotrichosis in a native of the Republic of South Africa. In vivo, this variety is characterized by its large, thick walled, hyaline tissue forms that range from 15-20 µm in diameter (Fig. 1a). Some of these cells multiply by a budding process, while others replicate by fission. In experimentally infected guinea pigs and mice, large, spherical mother cells with multiple buds are found mixed with those that have single buds. Most striking are the large cells with "eyeglass" configurations that result when septate cells remain joined following dissolution of one side of the parental cell wall. The *luriei* variety of S. schenckii thus differs significantly from S. schenckii var schenkii that has tissue form cells that are uniformally smaller, thinwalled and somewhat polymorphic. Most of the yeastlike cells of the classic variety are spherical to ellipsoid and measure 2-6 µm in diameter (Fig. 1b). Cylindrical forms, usually referred to as being cigar-shaped, measure 1-2 µm x 4-6 µm. All of these yeast-form cells typically reproduce by budding. *In vitro*, at both 25°C and 37°C, the two varieties of *S. schenckii* are indistinguishable from each other.

Discovery of the second case of fixed sporotrichosis with tissue forms of the *luriei* variety, 19 years after the first and only case was described, prompted this report.

CASE REPORT

Patient B.B., an 85-year-old, white man, born in Ponte dell'Olio, Piacenza, Italy, was admitted to the

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Figure 1a. - Single thick walled tissue form cell of *S. schenckii* var *luriei* shown at same magnification as the *schenckii* variety in Figure 1b: (GMS/H&E) x 1480.



Figure 1b. - Tissue form cells of *Sporothrix schenckii* var. schenckii include a budding and a single cell: (GMS) x 1480.

The excised tissue included the epidermis, and dermis, and portions of subcutaneous tissue. The epidermis revealed segmented hyperkeratosis and florid pseudoepitheliomatous hyperplasia (Fig. 2) with the formation of intraepidermal abscesses and transepidermal elimination channels (Fig. 3). The dermis contained a mixed suppurative and granulomatous inflammatory reaction accompanied by focal hemorrhage and intense vascularization by small blood vessels. A few eosinophils, lymphocytes

Histopathologic Findings

and plasma cells also were present.

Figure 2. - Cutaneous sporotrichosis caused by *Sporothrix* schenckii var. *luriei*. A hyperplastic epidermis overlies a mixed suppurative and granulomatous inflammatory infiltrate with scattered fungal elements in the dermis: (GMS/H&E) x 200.

Civil Hospital of Borgonovo, Piacenza, in July 1987. During the previous two months, he had gradually developed a raised nodule on the middle third of the exterior aspect of his lower right leg. The nodule was verrucose, with scattered crusts on its surface and in its greatest dimension measured 3.5 cm. The patient stated he had never traveled outside of Italy. After the physicians found no signs of bone involvement, the nodule, which was included in a strip of tissue 9.5 x 2.5 cm, was surgically excised. Since no one suspected the disease was caused by a fungus, the tissue was directly fixed in 10% neutral, buffered formalin and sent to the histology laboratory for study.

The day following surgical intervention, the patient developed chills and a fever of 39°C. During the next two days, the patient's condition deteriorated and he died before the histologic results could have provided insights for further studies or therapy.



Figure 3. - Intraepidermal microabscesses contain septate (arrows) and budding (blunt arrow) cells of *Sporothrix schenckii* var. *luriei*: (GMS/H&E) x 480.

Mixed with the dermal inflammatory elements, either extracellulary within microabscesses (Fig. 4) or intracellularly within epithelioid histiocytes, and multinucleated giant cells (Fig. 5) were numerous spherical, hyaline thick walled fungal cells that ranged from 10 to 20 µm in diameter. Fungal cells were also present in intra-epidermal abscesses and the hyperkeratotic layer of the epidermis. Some of the microabscesses contained small aggregates of fungal elements, many of which were morphologically atypical and degenerated. In the deep dermis were found individual and clustered degenerated fungal elements, together with intact fungal cells that clearly showed septations in either one or two planes. In addition, classical "eyeglass" cells, (Fig. 6) germinating yeast-like cells (Fig. 7), and short, frequently septate, branched hyphae were found. Some of the fungal cells were intimately surrounded by an intensely eosinophilic, periodic acid-Schiff positive material (Splendore-Hoeppli reaction) with either indistinct or spicular margins (26).

On observing the bizarre fungal elements present in the histologic sections, the possibility that they represented the tissue forms of *S. schenkii* var *luriei* was considered. This possibility was confirmed when deparaffinized tissue sections were stained with a fluorescent antibody (FA) reagent specific for *S. schenckii* (1, 17). The FA reagent stained the various



Figure 4. - A dermal abscess contains single cells and a dividing cell with septations in two planes (arrow): (GMS/H&E) x 480.



Figure 6. - Classical "eye glass" form of *S. schenckii* var. *luriei* formed by the incomplete separation of a septate cell and partial dissolution of the parental wall: (GMS/H&E) x 1480.



Figure 5. - Distorted thick walled, single and septate cells of *Sporothrix schenckii* var. *luriei*, 10 to 20 µm in diameter, are within epithelioid histiocytes and multinucleated giant cells of a dermal granuloma: (GMS/H&E) x 480.



Figure 7. - A germinated tissue form cell (arrow) and two septate cells (blunt arrow) of *Sporothrix schenckii* var. *luriei* embedded in a dermal abscess; (GMS/H&E) x 480.

fungal cells and hyphae intensely and thus confirming the diagnosis of the second case of sporotrichosis known to be caused by the *luriei* variety of *S. schenckii*.

DISCUSSION

In 1896 the first case of sporotrichosis came to medical attention when Benjamin Robinson Schenck studied the disease in a Maryland patient. He described the case in 1898 (30). Seven years later the first European case was diagnosed by Lucien de Beurmann and Louis Raymond at the Hopital Saint-Louis in Paris (4). In the ensuing nine years, de Beurmann and his colleague, Henri Gougerot, compiled data on 200 cases of sporotrichosis. From observing these cases, they described the disease in comprehensive detail in their historically valuable monograph, "Les Sporotrichose" (5).

In a manner, as yet unexplained, sporotrichosis had virtually disappeared throughout Europe by the end of the Great War (1914-1918) (29). That its low incidence and prevalence still persists is supported by a literature search that revealed only 44 cases reported from seven of the 35 European countries during the period of 1976-1988 (Table 1). Elsewhere in the world, sporotrichosis still flourishes in the Americas, especially in Latin America, as well as Africa and Asia

TABLE 1. - Documented European human cases ofsporotrichosis 1976-1988.

References	Countries	No. of Cases
16	Czechoslovakia	1
15, 19, 25	France	3
2, 6, 11, 12, 21, 23, 28, 33	Italy	14
18	Netherlands	1
10	Romania	1
3, 7 - 9, 13, 14, 20, 24, 31, 32	Spain	23
27	United Kingdom	1
Total	7	44

(29). The tissue form of the *luriei* variety of *S. schenckii* poses a challenge to histopathologists since it is strikingly different from the well-known tissue form of *S. schenckii* var *schenckii*. There are hints in the literature that one other case of sporotrichosis caused by the var *luriei* has occurred. As related by Ajello and Kaplan (1), a photomicrograph of a tissue section published by Mercadal-Peyri *et al.* in 1965 (22) depicted hyaline cells that resembled in size and configuration the *in vivo* "eyeglass" forms that characterize *S. schenckii* var. *luriei*. The Spanish

clinicians had diagnosed their patient's infection as cutaneous blastomycosis. A culture isolated from their patient's knee was identified as either a micrococcus or a torula. In all likelihood their isolate represented a contaminant. When awareness of the diagnostic tissue form of this variety becomes better known, *S. schenckii* var. *luriei* may be revealed to be less uncommon than it is currently assumed to be.

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