## RECURRENT LYMPHANGITIS OF THE LEG ASSOCIATED WITH DERMATOPHYTOSIS

## Report of 25 Consecutive Cases

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THE syndrome of recurrent lymphangitis, often resulting in permanent lymphedema, has been recognized for many years. Ochsner, Longacre, and Murray' gave a comprehensive discussion of this syndrome in 1940, and yet, even today, the clinical diagnosis of acute lymphangitis is frequently missed. Moreover, the cause of the recurrent attacks of lymphangitis is still not clear. Some authors<sup>2,3</sup> believe that recurrence is secondary to foci of infection in the body. Others<sup>4</sup> believe that recurrence is due to an acquired hypersensitivity of previously infected tissues. The majority of authors believe that usually there is some portal of entry for bacteria through the skin in order to cause the lymphangitis.

That there is more than a casual relationship between dermatophytosis (athlete's foot) and the repeated attacks of lymphangitis has been suggested by Allen and Ghormley,<sup>5</sup> by Sulzberger, Rostenberg, and Goetze,<sup>6</sup> and in several textbooks.<sup>7-11</sup> It is the purpose of this paper to discuss 25 consecutive cases of recurrent lymphangitis associated with dermatophytosis treated at the Cleveland Clinic in the years 1950 through 1958, and the clinical course subsequent to treatment.

## Clinical Signs and Symptoms

The attacks of lymphangitis are characteristic in the acuteness of their onset and the severity of the systemic reaction. They usually occur suddenly, without warning, and within a few hours the symptoms are extremely severe. Occasionally there is a dull pain in the affected extremity, which rapidly becomes acute. A severe, shaking chill with fever as high as 106 F. usually initiates the attack. Associated symptoms include malaise, anorexia, headache, nausea and vomiting. The regional lymph nodes of the extremity often become swollen and tender. On the affected limb a small red area may appear, which rapidly spreads so that soon a considerable part of the extremity is red, swollen, and tender. Frequently there are red streaks running along the course of the lymphatic vessel. A fever persists from a few hours to two or three days. The chills may recur every few hours. The duration of the attacks varies from 2 to 14 days, but there is a characteristic, quick response to penicillin or to penicillin-like drugs. Some residual swelling may persist after all the clinical signs of acute inflammation have disappeared.

The intervals between attacks vary greatly. They may occur as frequently as once a week, but usually they are several months apart. As the disease advances and the lymphedema worsens, the periods between attacks usually become shorter.

Single attacks of lymphangitis do not as a rule result in persistent lymphedema. However, with each subsequent attack the edema becomes more severe and even-

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tually becomes irreversible. Thus it is important not to ignore the early attack, but to treat it and its source quickly.

Cases of acute lymphangitis with cellulitis are most frequently misdiagnosed as thrombophlebitis, though the differential diagnosis is usually quite easy to make. The distinguishing characteristics are summarized in *Table 1*.

| Differential<br>features         | Lymphangitis<br>with cellulitis | Superficial throm-<br>bophlebitis | Iliofemoral throm-<br>bophlebitis |  |
|----------------------------------|---------------------------------|-----------------------------------|-----------------------------------|--|
| Onset                            | Sudden                          | Gradual                           | Gradual                           |  |
| Fever                            | High—up to<br>106 F.            | Mild—99 to<br>101 F.              | Rarely more<br>than 102 F.        |  |
| Chills                           | Common                          | Rare                              | Rare                              |  |
| Lymphadenopathy                  | Common                          | Rare                              | Rare                              |  |
| Portal of entry<br>for infection | Usual                           | Rare                              | Rare                              |  |
| Palpable, in-<br>flamed vein     |                                 |                                   | Common                            |  |
| Red streaking<br>of skin         | Common                          | Common                            | None                              |  |
| Size of limb                     | Enlarged                        | Normal                            | Enlarged                          |  |
| Superficial veins                | cial veins Normal Norm          |                                   | Distended                         |  |
| Skin tempera-<br>ture of limb    | High                            | High                              | Usually normal                    |  |
| Response to penicillin           | Rapid                           | None                              | None                              |  |

**Table 1.**—Differential diagnosis of inflammation of the leg: lymphangitis with cellulitis, superficial thrombophlebitis, and iliofemoral thrombophlebitis

# Clinical Data

Twenty-five cases of recurrent lymphangitis associated with dermatophytosis have been treated at the Cleveland Clinic in the years 1950 through 1958 (*Table 2*). There are from one to eight year follow-up records on 20 patients. One patient died, and four patients were lost to follow-up. The age range was 22 years to 67 years. There were 9 women and 16 men; they had from 2 to 32 attacks of lymphangitis in from 1 to 30 years. The most unfortunate patient had 30 attacks within three years.

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Of 16 patients who had been given diagnoses before being examined here, 13 patients were told that they had either thrombophlebitis or recurrent thrombophlebitis. Three patients were thought to have cellulitis. A diagnosis of rheumatic fever was considered in two patients, osteomyelitis in one patient.

One patient (no. 13, Table 2) apparently had a flare-up of nephritis with gross

| Table 2.—Data on 25 consecutive patients who had recurrent lymphangiti | s of the leg with |
|--|-------------------|
| dermatophytosis treated at Cleveland Clinic (1950 through 19           | )58)              |

| Case<br>no. | Age,<br>years | Sex | Attacks, to-<br>tal number | Interval<br>since first<br>attack, years | Attacks since<br>treatment,<br>number | Follow-up,<br>years | Recurrent fungu<br>since treatment |
|-------------|---------------|-----|----------------------------|--|---------------------------------------|---------------------|------------------------------------|
| 1           | 55            | F   | 3                          | 1  | 1                                     | 5                   | Yes                                |
| 2           | 46            | Μ   | 30                         | 3  | 1                                     | 4                   | Yes                                |
| 3           | 41            | М   | 15                         | 1  | 5                                     | 4                   | Yes                                |
| 4           | 44            | F   | 4                          | 10                                       | 1                                     | 5                   | Yes                                |
| 5           | 22            | Μ   | 24                         | 12                                       | 7                                     | 7                   | Yes                                |
| 6           | 59            | Μ   | 6                          | 30                                       | 0                                     | 4                   | No                                 |
| 7           | 38            | F   | 3                          | 3  | 0                                     | 4                   | No                                 |
| 8           | 63            | М   | 14                         | 4  | 0                                     | 31/2                | No                                 |
| 9           | 33            | М   | 2                          | 1  | 0                                     | 7                   | No                                 |
| 10          | 34            | Μ   | 16                         | 7  | 0                                     | 4                   | No                                 |
| 11          | 31            | М   | 13                         | 2  | 0                                     | 1¾                  | No                                 |
| 12          | 37            | F   | 26                         | 10                                       | 0                                     | 21/2                | No                                 |
| 13          | 33            | F   | ``Many"                    | 15                                       | 0                                     | 4                   | No                                 |
| 14          | 57            | М   | 5                          | 5  | 0                                     | 7                   | No                                 |
| 15          | 31            | М   | 7                          | 9  | 0                                     | 6                   | No                                 |
| 16          | 53            | М   | 4                          | 8  | 0                                     | 8                   | No                                 |
| 17          | 67            | М   | 5                          | 5  | 0                                     | 5                   | No                                 |
| 18          | 37            | F   | 11                         | 2  | 0                                     | 1                   | No                                 |
| 19          | 30            | М   | 2                          | 1  | 0                                     | 1                   | No                                 |
| 20          | 55            | F   | 20                         | 2  | 0                                     | 6                   | No                                 |
| 21          | 63            | F   | 4                          | 6  | Deceased                              |                     |                                    |
| 22          | 29            | М   | 2                          | 1  |                                       |                     |                                    |
| 23          | 33            | F   | 5                          | 20                                       | Lost to follow-up                     |                     |                                    |
| 24          | 30            | М   | "Several"                  | 17                                       |                                       |                     |                                    |
| 25          | 22            | М   | "Several"                  | 2)                                       |                                       |                     |                                    |

hematuria during each attack of lymphangitis, an association that strengthened the belief that the lymphangitis was of streptococcal origin.

All patients had dermatophytosis proved by potassium hydroxide preparations

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of skin scrapings. All were treated with undecylenic acid-zinc undecylenate ointment, sometimes in combination with Whitfield's ointment, in the manner outlined under *Prevention of future attacks*.

Mild to moderate lymphedema was noted in 12 patients when first examined, and two had severe lymphedema. In the follow-up questionnaires, 10 patients reported some swelling of the legs and feet, which disappeared at night. Only three patients stated that they had swelling that persisted day and night.

A striking fact is that, of the 20 patients who were followed, 15 have had no further attacks. These patients stated that they have been able to keep their feet free of dermatophytosis. Of the five patients who have continued to have attacks, all still have dermatophytosis.

## Treatment

Treatment of the acute attack. Acute attacks of lymphangitis will usually subside spontaneously within 4 to 14 days; however, the response to penicillin and similar antibiotics is usually quite dramatic within one to three days. Additional measures to be taken include bed rest, elevation of the affected extremity, and the application of warm, moist packs.

*Prevention of future attacks.* In addition to the specific therapy in acute lymphangitis, between attacks it is important that edema of the limb be controlled as much as possible because the edematous tissue is highly susceptible to infection. A properly fitted elastic stocking to control the swelling in the leg should be worn at all times when the patient is up and about. The affected leg should be elevated at night and whenever possible during the day. An occasional course of oral diuretics may be needed. With more resistant edema, pneumatic massage and precise fitting with a prestressed, elastic sheath as outlined by Brush and associates,<sup>12</sup> and by Britton<sup>13</sup> may be necessary. This control of edema can be a major factor in preventing future attacks.

The sources of infection and reinfection must be eradicated. This is especially necessary when there is dermatophytosis. An ointment containing undecylenic acid and zinc undecylenate is usually prescribed to be applied between the toes twice daily until all obvious infection has cleared, usually for about two weeks. After the skin has healed, the ointment is applied once each week, and a powder containing the same ingredients is dusted in the shoes and the stockings about three times weekly. Patients are advised to dry well between the toes after bathing and to wear clean stockings daily. This treatment should be followed indefinitely. For the more resistant cases, especially those in which there is onychomycosis, the new oral antifungal preparation griseofulvin is proving especially effective.

## Discussion

The patient who has repeated attacks of a red-hot, painful, swelling of the leg can present quite a problem in diagnosis to the physician who is unaware of the clinical entity of recurrent lymphangitis. Idiopathic recurrent thrombophlebitis

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may be strongly suspected, and a fruitless, expensive search may be made for occult carcinoma. A needless, potentially dangerous course of prophylactic anticoagulants may be tried. A frustrating situation occurs when the patient continues to have repeated attacks despite the application of the best diagnostic and therapeutic measures known to the physician.

It has long been recognized that there is usually some break in the skin to serve as a portal of entry for bacteria in cases of lymphangitis. A break such as a wound, cut, ulcer, abrasion, contusion, scratch, hangnail, pinprick, or vesicle, usually can be found if a careful search is made. Even minor abrasions with the slightest wound of the corium may tear open lymph vessels and permit direct entry of bacteria. Dermatophytosis with moist, warm, macerated skin, and fissures between the toes provides an ideal medium and entry for bacteria. Also, because of its chronic nature, it provides a portal of entry for repeated attacks of the bacteria on the lymphatic system over periods of months and years.

When bacteria enter the lymphatic vessels a spreading, obliterative lymphangitis develops. Attempts to find the causal organism are usually unsuccessful. It is said that in 1892 Sabouraud first demonstrated the presence of the streptococcus organism in patients with recurrent lymphangitis.<sup>1</sup> In 1930, Suarez<sup>14</sup> reported a study of 60 cases of recurrent lymphangitis, and cultures obtained from the specimens of subcutaneous tissue in 50 patients were negative in all but two patients; streptococci were found in both of them. The streptococcus often can be isolated from fissures of the skin associated with dermatophytosis.

The lymphangitis causes thrombosis and thereby occlusion of the lymph vessels. The inflammatory reaction causes excess accumulation of proteins in the interstitial fluid. This storing up of proteins sets up a vicious cycle, for the flow of lymph is impeded and still more proteins accumulate. The protein and other products of inflammation are ideal for the growth of fibroblasts and the production of scar tissue. Drinker, Field, Ward, and Lyons<sup>15</sup> demonstrated that the lymphedema fluid itself is a rich medium for bacteria, and greatly increases the chances for future attacks of lymphangitis. With each subsequent attack the affected area becomes larger and there is thickening and scarring of the subcutaneous tissue; eventually typical elephantiasis with massive limbs may result. It is important to recognize this clinical entity of recurrent lymphangitis in its early stages before much permanent damage has been done.

### Summary

Recurrent lymphangitis is a well-defined but too frequently misdiagnosed clinical entity. It is characterized by sudden, repeated attacks of a red-hot, painful, swelling of the leg. Proper diagnosis is essential. Treatment includes bed rest, elevation of the leg, elastic-sheath covering of the leg if needed. When dermatophytosis is also present, appropriate local therapy and, if indicated, griseofulvin are used. This therapy must be instituted early in the course of the disease to preclude massive edema and elephantiasis.

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The data of 25 consecutive cases of recurrent lymphangitis associated with dermatophytosis have been presented; the clinical course after treatment is included. After the dermatophytosis cleared and the edema was controlled, no future attacks occurred in 15 patients. Recurrent attacks of lymphangitis continued in five patients who were unable or were unwilling to control the dermatophytosis.

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