

OCCURRENCE OF ERYTHEMA CHRONICUM MIGRANS AND LYME DISEASE AMONG CHILDREN IN TWO NONCONTIGUOUS CONNECTICUT COUNTIES

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In May–November 1977 erythema chronicum migrans or Lyme arthritis occurred in at least 12 children in New London County, Connecticut, east of the original 3-town epidemic focus. The attack rate (0.15 cases per 1,000 persons under 18) was considerably lower than within the focus itself but similar to the rate in towns west of the focus. No cases were identified in Litchfield County in northwest Connecticut. Observed rates of Lyme disease varied markedly within Connecticut in 1977.

Investigators from Yale and the Connecticut State Department of Health have described in detail the clinical and epidemiologic features of Lyme disease—an inflammatory disorder primarily of the skin, joints, nervous system, and heart (1–3). Studies in Connecticut (1–5) provide strong evidence that the bite of ticks now known as *Ixodes dammini* initiates the syndrome, often with the unusual dermatologic manifestation of erythema chronicum migrans (ECM)—i.e., redness that expands centrifugally for up to

several weeks to form an annulus, often many centimeters in diameter. No etiologic agent or diagnostic laboratory test has been found. Since its first description in 1976 in Lyme, Old Lyme, and East Haddam, Connecticut, examples of probable cases of Lyme disease have come from elsewhere on the East Coast, from Nantucket Sound to as far south as the Baltimore area, and from Wisconsin, California, and Oregon (6,7). However, at the time this survey was organized, no systematic epidemiologic studies had documented the frequency of cases in areas outside of the towns where the disease was first recognized.

The study reported here, undertaken in the warm season of 1977, was an effort to complement the investigation by Steere and his colleagues (3) of the occurrence of Lyme disease. It extended epidemiologic observations beyond the epidemic focus and assessed the proportion of inflammatory childhood arthritis attributable to Lyme disease in those areas during the study period.

MATERIALS AND METHODS

We searched for ECM and arthritis with onset in a 7-month period in children residing in 2 areas of Connecticut. All of New London County east of Lyme, Old Lyme, and East Haddam (Figure 1) was chosen because sporadic cases of Lyme disease had been noted in residents there. Lyme and Old Lyme were excluded because they were already under intensive study. Litchfield County was selected because it is not contiguous with New London County, because neither cases of Lyme disease nor *I dammini* had been seen there previously, and because it has a nonurbanized, woodland environment and self-contained provision of primary medical care.

We confined the survey to children under 18 years for several reasons. The original report (1) on Lyme disease

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suggested that most of the people affected were children. It appeared, too, that in this survey children would have a lower frequency of arthritis and fewer forms of arthritis to differentiate. In addition, they are generally more likely than adults to be given medical care for less florid illness and to be seen by a relatively easily defined subset of health professionals.

There was considerable publicity about Lyme disease. To provide all participants with common knowledge of the condition, immediately before the survey period the Connecticut State Department of Health supplied a description of Lyme disease to all physicians licensed in the state. The 430 internists, dermatologists, pediatricians, family and general practitioners, orthopedic surgeons, and doctors of osteopathy serving the 2 survey areas also each received a color photograph of the lesion of ECM and an explanation of the study as part of an invitation to participate. We later determined that 341 physicians were eligible, i.e., actively engaged in an appropriate kind and place of practice. Of those, all but 30 (8%) cooperated in the survey.

A trained assistant from the Connecticut State Department of Health telephoned each participating physician's office 5 times in the period May 1–December 13, 1977 to elicit a report on any patient with ECM or nontraumatic arthritis with onset after May 1. Using a questionnaire designed for this study, the assistant obtained a clinical and environmental exposure history from the physician or parent of each child with a suspected case. She also obtained available followup information on cases reported earlier in the survey. Investigators from Yale forwarded data on all appropriate cases that came to their attention.

Three types of illness were reported: 1) ECM without joint involvement, 2) ECM with joint involvement, and 3) inflammatory joint disease without ECM. Lyme disease, comprising the first 2 types, was defined as ECM with or without pain and/or swelling that began in the period May 1–November 30, 1977 in a resident of the survey area who was under 18 years of age.

Population estimates were derived from 1977 figures in the state of Connecticut Register and Manual, 1978 (8). Although the total state population increased between 1970 and 1977, the proportion under 18 years of age decreased. Adjustments for those changes produced an estimate of 77,593 residents under 18 in New London County, excluding Lyme and Old Lyme, and 50,642 under 18 in Litchfield County.

RESULTS

Twelve cases satisfying our epidemiologic definition of Lyme disease were found in the New London County survey area (Table 1, Figure 1). The 7-month incidence rate was 0.15 per 1,000 (95% confidence interval, 0.06–0.24 per 1,000). No cases were reported from Litchfield County. This difference was statistically significant ($P = 0.0025$, binomial test). There is a suggestion that the rate was lower in the northern half of the New London County survey area (0.04 per 1,000) than in the southern half (0.2 per 1,000); howev-

er, the numbers of cases were too small to be significant ($P = 0.09$, binomial test). There were no important differences between patients in this study and those previously described (1–3) in terms of age, sex, date of onset, or the proportion with brief and recurrent joint inflammation, knee involvement, neurologic symptoms, or tick bite or skin contact in the previous 90 days.

Five patients had inflammatory arthritis without antecedent ECM; all 5 of these patients were residents of New London County. Three had a clinical presentation and course somewhat atypical for Lyme disease and were diagnosed as having juvenile rheumatoid arthritis by a physician. Information obtained in those cases during the survey and the 2-year follow-up period was insufficient to establish a diagnosis according to criteria for the arthritis of Lyme disease (3), juvenile rheumatoid arthritis (9), or other rheumatic diseases.

DISCUSSION

Our 1977 survey disclosed a difference that could not easily be attributed to chance between the rate of Lyme disease in children residing in the New London County survey area (0.15 per 1,000) and the null incidence in Litchfield County. We also doubt that any biases involved in recognizing and reporting Lyme disease could account for the different rates in New London and Litchfield counties. Repeated public accounts of the illness, a widely disseminated state health department newsletter, the direct invitation to physicians to participate in the survey, and repeated telephone contact with physicians' offices should have adequately reinforced awareness of the illness in both survey areas. The lesion is distinctive enough that few physicians who are so reminded of it are likely to mistake it for another condition. Participation by physicians in both survey areas was nearly complete.

Other possible sources of error in our estimates seem worth mentioning. First, 1 girl from the town of New London had traveled briefly to East Haddam very early in the tick season and could have acquired the illness there. Second, only patients with ECM were included in our estimates. Although it has a highly specific dermatologic sign, ECM can be asymptomatic. A small proportion of affected patients either do not have or do not remember the skin lesion and seek medical attention only for rheumatic or neurologic complaints (3). Third, 5 individuals in the New London County study area had arthritis without ECM; 2 cases were indistinguishable from Lyme disease but

Table 1. Demographic and clinical data of 12 patients with Lyme disease, New London County, Connecticut, May–November 1977

| Case | Age | Sex | Town | Date of onset | ECM | Neurologic symptoms | Duration of first attack of joint symptoms (days) | Attacks 5/1–12/31 | Other compatible symptoms* | Lab† | Tick contact |
|------|-----|-----|---------------|---------------|-----|---------------------|---|-------------------|----------------------------|-----------|--------------|
| 1 | 17 | F | N. Stonington | June | + | – | – | 1 | + | ESR | – |
| 2 | 10 | F | Stonington | June | + | + | – | 1 | – | ESR | ? |
| 3 | 13 | M | Salem | Aug | + | – | – | 1 | – | – | – |
| 4 | 4 | M | E. Lyme | May | + | + | – | 1 | + | ESR | – |
| 5 | 16 | F | New London | May | + | + | – | 2 | + | ESR, Cryo | – |
| 6 | 10 | F | Stonington | June | + | + | 49 | 2 | + | – | + |
| 7 | 14 | F | E. Lyme | June | + | + | 77 | 2 | + | ESR | + |
| 8 | 8 | M | Waterford | July | + | – | 1.5 | 2 | – | ESR | – |
| 9 | 9 | M | New London | May | + | + | 13 | 2 | + | Cryo | + |
| 10 | 7 | F | E. Lyme | July | + | – | 1 | 2 | – | ESR, Cryo | – |
| 11 | 12 | F | Norwich | June | + | + | 75 | Multiple, brief | + | ESR | + |
| 12 | 12 | F | Montville | June | + | + | 73 | Multiple, brief | + | – | – |

* Two or more of the following within 10 days of onset of erythema chronicum migrans (ECM) or joint symptoms: fever, chills, malaise, photosensitivity, pharyngitis, myalgia, anorexia, nausea or vomiting, palpitation, lymphadenopathy, abdominal pain.
 † ESR = abnormal erythrocyte sedimentation rate; Cryo = cryoglobulin present.

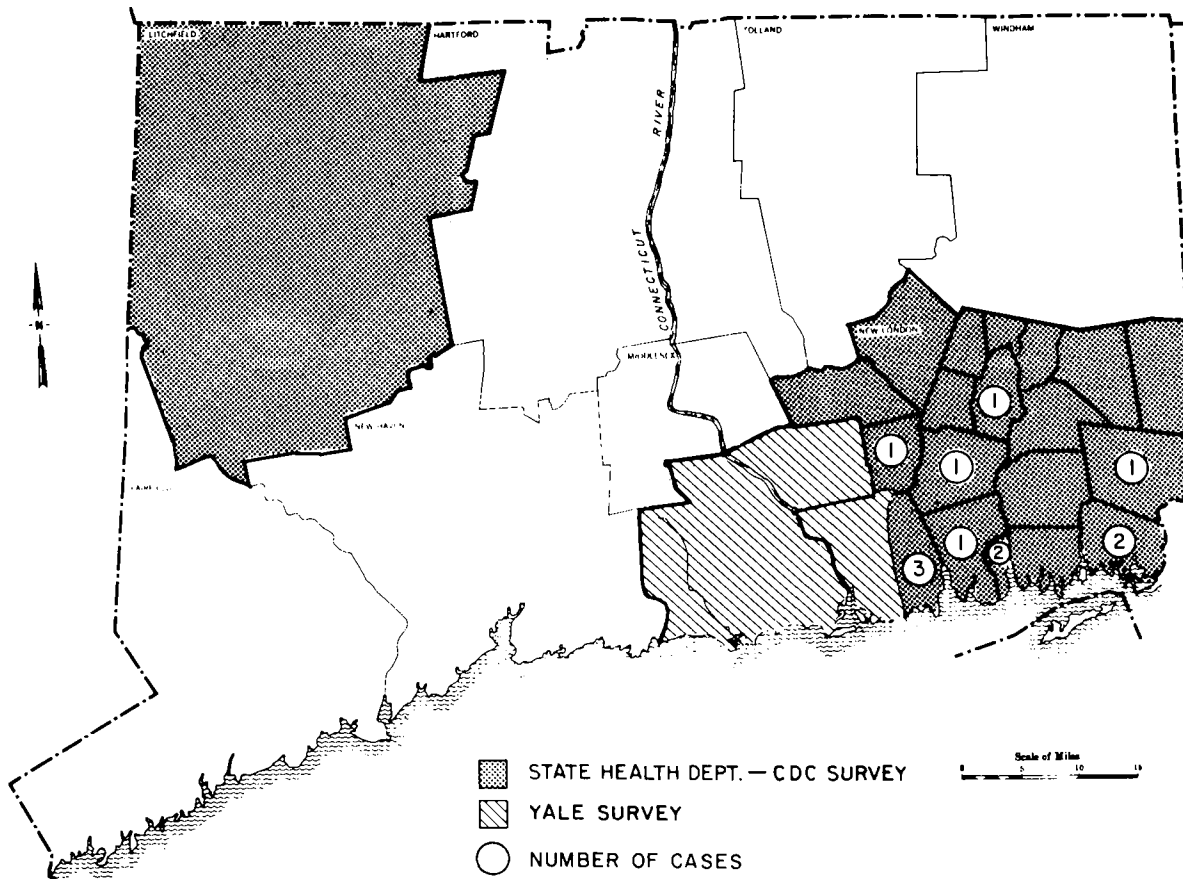


Figure 1. Study areas for active surveillance of Lyme disease, Connecticut, May–November, 1977.

were not counted in the case group because they did not meet all criteria applied by Steere and coworkers (3). The 3 diagnosed as juvenile rheumatoid arthritis could have been Lyme disease although they were clinically atypical. Moreover, on the basis of published prevalence figures (10) and the average duration of juvenile rheumatoid arthritis, we would have anticipated 1 to 6 new cases in the New London County and none to 4 in Litchfield County in the 7-month interval. Fourth, a rare case seen only by a school nurse or not seen by a health professional at all may have been overlooked. Finally, the circumscribed surveillance period may have excluded that small proportion of patients who had trivial symptoms during the 7-month interval but developed illness for which they sought medical attention only after November, when our survey ended.

Although our survey methods differed from those of Steere and his coworkers, the rate we estimated for eastern New London County (0.15 per 1,000) was on the same order of magnitude as the rate based on the number of cases under 18 seen by the Yale group from west of the epidemic focus (0.21 per 1,000) (11). Conversely, the rate estimate for eastern New London County (0.15 per 1,000) was nearly 30 times lower than the estimate for the population under 18 from Lyme, Old Lyme, and East Haddam (4.4 per 1,000).

We do not believe that the methodologic problems cited undermine the 2 basic conclusions: that Lyme disease accounted for most of the inflammatory joint disease seen in children during the warmer months of 1977 in eastern New London County, and that the incidence of the disease in 1977 showed considerable geographic variability in Connecticut. The incidence of the disease appears to have been highest in the vicinity of the original epidemic focus, lower in eastern New London County but roughly equal to the rate in the Yale study area west of the Connecticut River, perhaps less frequent in the inland northern half than in coastal southern half of New London County, and undetectable in Litchfield County.

Evidence to date (6) suggests that Lyme disease occurs where the newly designated tick, *I dammini*, or ticks of the *I ricinus* complex are found; moreover, in 1 study the incidence rates seemed to vary with the tick population density (4). Although we know of no active search for *I dammini* in Litchfield County, this tick species has been documented only within areas of Connecticut closer to the Atlantic Coast. Therefore,

the most likely explanation for our failure to find any Lyme disease in Litchfield County is that the tick vector was not present there. Other factors such as the density of the nonhuman host populations and the intensity and duration of their carriage of the etiologic agent may also be important in explaining the great variability in incidence of Lyme disease within the habitat of *I dammini*.

There is another fact whose epidemiologic significance for Lyme disease is unclear. *I dammini* also transmits *Babesia microti*, the protozoan etiologic agent of human babesiosis, a rare infectious disease affecting primarily older, often splenectomized adults. In contrast to Lyme disease, it has been recognized only on Cape Cod, its offshore islands, and Shelter Island in Long Island Sound (12). It has not been reported in Connecticut or the other parts of the Northeast, Wisconsin, or the Pacific Northwest where Lyme disease has been seen. The discrepancies in geographic distribution may be due to chance or to misdiagnosis of babesiosis but more likely reflect differences between *B microti* and the unknown etiologic agent of Lyme disease in their intrinsic biology or mode of transmission. While these and other unanswered questions await the discovery of the Lyme disease agent, study of the occurrence of Lyme disease and babesiosis, their nonhuman reservoirs, and their vectors could be fruitful as a basis for controlling the diseases even before an agent is identified.

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