

Epidemic Neuromyasthenia, An Outbreak In Punta Gorda, Florida

An Illness Resembling Iceland Disease

WALTER B. CLEMENT, M.D.*, PUNTA GORDA, DONALD A. HENDERSON, M.D.†, ATLANTA, GA., JOSEPH W. LAWRENCE, M.D.‡, ARCADIA, AND JAMES O. BOND, M.D.§, JACKSONVILLE

In the spring of 1956, there appeared in the south Florida community of Punta Gorda a bizarre disease previously unknown to those of us there. The illness was characterized principally by pronounced fatigue; pain in the head, neck, and extremities; nausea and vomiting; dizziness; paresthesias; emotional lability and depression; impairment of memory; and a protracted, relapsing course.

A major epidemic composed of cases of a strikingly similar nature was observed in Tallahassee in the fall of 1954.¹ Additional outbreaks in Florida were reported from Bradenton in the fall of 1955,² and from Lakeland in the fall of 1952.³ In recent years, reports of similar epidemics have come from many parts of the world⁴ and from points in this country as divergent as southern California⁵ and upper New York state.⁶ In England, similar illnesses have been commonly called "benign myalgic encephalomyelitis,"⁷ while in this country, the term "Iceland disease" has found greater favor.

The high attack rates experienced among populations affected and the protracted debilitating course of the illness serve to produce a situation commanding the interest and concern of the practicing physician.

Punta Gorda Outbreak

In retrospect, the epidemic in Punta Gorda appears to have commenced in February of 1956. Cases initially were infrequent. Anxiety and depression evidenced by the patients, the non-specific symptomatology, the multitude of symptoms and contrasting relative absence of positive physical and laboratory findings, and the frequent occurrence of cases among middle-aged women

*Private Physician, Punta Gorda.

†Assistant to the Chief, Epidemiology Branch, Communicable Disease Center, Public Health Service, Atlanta, Ga.

‡Health Officer, Charlotte County Health Department, Arcadia.

§State Epidemiologist, Florida State Board of Health, Jacksonville.

Read before the Florida Health Officers' Society, Twelfth Annual Meeting, Hollywood, May 5, 1957.

readily served to obscure diagnosis and identification of the existence of the epidemic in its initial phases. "Neurosis" was the usual case diagnosis, not inconsistent with the findings and an initial diagnosis commonly made in other epidemics.

By late April, the appearance of larger numbers of cases and the failure of recovery among earlier cases produced increasing concern in Punta Gorda and the recognition of the need for a major community-wide study. Assistance was sought, and in May investigations were initiated by the Florida State Board of Health and the Charlotte County Health Department. Additional support was subsequently provided by the Public Health Service's Communicable Disease Center.

Cases continued to occur throughout May with the peak occurrence of cases coming during the first week. The appearance of new cases fell rapidly thereafter and ceased by the first week in June.

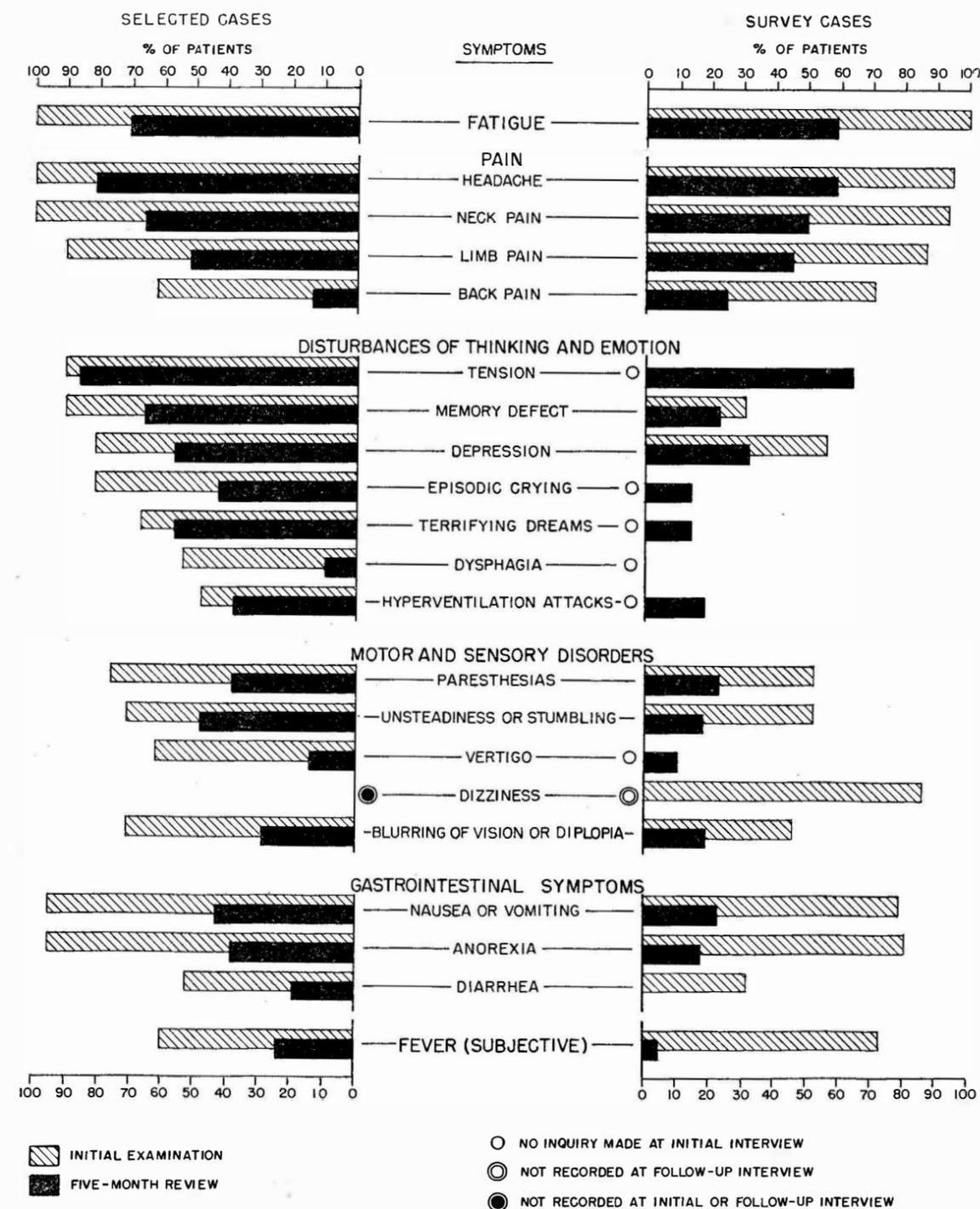
Symptoms

Initial studies were focused on a better definition of the illness. Twenty-one patients who reported being currently ill were selected for detailed clinical and laboratory study. Seventeen of them were female; all were white. They ranged in age from 14 to 60 years.

The onset in most was insidious, marked by gradually increasing fatigue, headache, and pain in the neck, back, and extremities. Commonly, dizziness, emotional tension, depression, and some mental confusion commenced soon after the onset. In about half, there was a pronounced, abrupt exacerbation of symptoms between one and four weeks after the onset. The progressive course of the disease is reflected by the fact that, among 19 of this group who became bedridden, the median time between onset and confinement to bed was 19 days.

Figure 1 depicts the relative frequency of the more common symptoms. Fatigue was the most incapacitating symptom in terms of frequency and

FIGURE 1
FREQUENCY OF OCCURRENCE OF SYMPTOMS



persistence. Headache was, in general, poorly localized, nonthrobbing in character, and little relieved by analgesics. Severe occipital or suboccipital pain radiating toward the frontal area was common. Limb and back pains were similar in nature in each of the areas, being described as aching in quality, "deep" and "diffuse."

Emotional changes were particularly notable. Anxiety and depression were reported by almost all patients. Episodes of crying without provocation and frequent terrifying dreams were common. Difficulty in recalling recent events was reported by many. During the acute phase particularly, a number of patients noted a tendency to transpose letters when writing or words when talking. Two symptoms customarily referred to as neurotic were encountered with unusual frequency. These were hyperventilation attacks and dysphagia without regurgitation.

Paresthesias, assuming the form of numbness and tingling of the extremities, were reported by most patients. Paresthetic areas were patchy, however, and not infrequently shifted from week to week. No nerve or root pattern for these could be discerned.

Unsteadiness or stumbling was primarily subjective and not particularly evident to observers.

Anorexia and nausea, often with vomiting, occurred in almost all patients. Vomiting in a few became severe enough to require intravenous fluid therapy. In many cases in which there was severe vomiting, patients reported symptoms consistent with vertigo.

Feverishness was reported by 13 of the 21 patients. On but five occasions were readings in excess of 100 F. noted. The temperature elevations rarely exceeded one day; the highest recorded temperature was 102 F. Menstrual irregularities, irregular periods or amenorrhea, were common.

Although emotional changes were overtly evident, objective physical findings in the patients were wholly disproportionate to the severity of their illnesses. A sensation of neck "tightness" resembling a mild nuchal rigidity was experienced by most patients when the neck was fully flexed. Reflexes were equal bilaterally although sometimes accentuated.

Mild to moderate impairment of touch, pain, and temperature in varying combinations were present in 12 patients. Distribution of the abnormalities, however, did not conform to the distribution of root or peripheral nerve zones.

Focal muscle tenderness, that is, areas about 5 to 8 cm. in diameter, were present in nine patients. A mild paresis was present in two although many more complained of real weakness. A Romberg sign was noted in five.

Laboratory studies revealed little. White blood cell counts and cerebrospinal fluid examinations performed were normal. Stools, throat washings, blood clots, and spinal fluids were tested by inoculation of suckling mice, HeLa, monkey kidney, and human liver tissue culture systems at the Virus and Rickettsia Laboratory, Communicable Disease Center. Despite two and three blind passages with many of the specimens, no agent was isolated. Bacteriologic cultures of stool specimens were negative. Paired serums from 12 patients were tested for antibodies to the arthropod-borne encephalitides, lymphocytic choriomeningitis, brucella and leptospira. These were all negative. Heterophil determinations were also negative.

From study of these 21 patients, certain problems became apparent. We appeared to be confronted with a disease of protean symptomatology with relatively few physical findings. There apparently was no individual symptom or constellation of symptoms specific enough to permit conclusive case diagnosis, and most perplexing was the observation that cases in less severe form blended increasingly with psychoneurotic illness.

Epidemiologic Characteristics of More Severe Cases

Recognizing then that for epidemiologic study, minor cases would necessarily have to be discarded, we formulated criteria which in this outbreak would include the more pronounced cases and more certainly exclude psychoneurotic illness. The criteria were as follows:

1. A definite change in physical and emotional status indicating an onset of illness during the epidemic period
2. Illness of seven or more days
3. Presence of at least six of the common symptoms

The epidemiologic characteristics to be presented are descriptive then only of the more severe cases.

A house-to-house survey was organized, in which half the population of Punta Gorda was sampled. Sixty-two cases were uncovered, which met the noted criteria. From the survey, it was apparent that at least 120 relatively severe cases had occurred in addition to a substantial number

of less severe cases. The comparative frequency of symptoms for this group as compared to the selected case group is included in figure 1. In general, the pattern of occurrence of symptoms is similar in the two groups although the frequency is somewhat reduced in the survey group, indicating an over-all less severe illness.

In table 1 are shown the age-specific attack rates for the survey cases. Notably absent are cases among children under 10 years of age. Our youngest patient actually was 12 years old. Despite intensive questioning of parents, we were able to uncover little illness of even a minor nature among children during the epidemic period. The attack rates for Negroes are based on small numbers and do not deserve further comment. Attack rates for white females are relatively constant through age 69. The males show a relatively similar rate through age 49, after which there is a substantial decline.

Despite the rather similar rates among males and females under the age of 50, it was the impression of all that the disease, though present, was less severe in the males.

Follow-Up Study

Five months after our initial visit to Punta Gorda, we interviewed and re-examined the same 21 patients initially intensively studied. They had shown slow but definite improvement interrupted by a series of exacerbations. Recurrence of symptoms seemed to be associated particularly with excessive fatigue or occurred with the onset of the menstrual period. Anxiety, depression, fatigue, and insomnia were most disabling of the symptoms persisting. A random sample of the survey group was similarly interviewed. This group was, on the whole, less disabled but experienced a similar persistent pattern of symptoms.

The emotional and physical debility evidenced by those in Punta Gorda six months after the onset of their illness was distressing. But two of 21 experienced asymptomatic days by the sixth month of illness. Five of this group had been confined to bed for one or more days as late as the sixth month following onset, two of these being confined to bed for the entire month. In over 40 per cent of the total patient days during the sixth month definite restriction of activity was necessitated because of symptoms. Additional follow-up studies on these patients would be of considerable interest.

Table 1.—Attack Rates by Age, Sex, and Race House-to-House Survey Punta Gorda, Florida

Age Group	White Female			White Male			Negro Female			Negro Male			Total		
	Cases	Pop.	Attack Rate (%)	Cases	Pop.	Attack Rate (%)	Cases	Pop.	Attack Rate (%)	Cases	Pop.	Attack Rate (%)	Cases	Pop.	Attack Rate (%)
0-9	0	51	—	0	62	—	0	19	—	0	18	—	0	15	—
10-19	6	56	10.7	5	46	10.9	0	18	—	0	23	—	11	143	7.7
20-49	15	131	11.5	11	121	9.1	4	36	11.1	1	26	3.8	31	314	9.9
50-69	10	110	9.1	3	106	2.8	4	25	16.0	0	17	—	17	258	6.6
70 & over	2	35	5.7	0	45	—	1	8	12.5	0	5	—	3	93	3.2
Unknown	0	16	—	0	23	—	0	8	—	0	5	—	0	52	—
	33	399	8.3	19	403	4.7	9	114	7.9	1	94	1.1	62	1,010	6.1

Many possible vehicles of transmission were investigated, including water, milk, various foods, cosmetics, soaps, and insecticides. None could be incriminated. Mosquitoes were reported as absent from the community until an influx on April 21.

Geographically, cases appeared to be limited to Punta Gorda and the rural area within a nine mile radius. Discussions with physicians in neighboring towns uncovered no cases in the adjacent communities.

Because of several reports which have appeared of outbreaks of a similar disease entity among hospital personnel, a survey was made of medical and allied personnel in Punta Gorda. This group constituted 38 persons in all. Applying the same case definition criteria to this group as were applied in the survey, 16 cases were uncovered. The attack rate for this group was 42 per cent, far in excess of that experienced by the town as a whole.

Discussion

The studies outlined illustrate certain of the difficulties encountered in the study of this disease entity. With no deaths occurring during this epidemic and none recorded in other epidemic situations, a pathophysiologic explanation for the symptoms remains obscure. There are no classical signs or symptoms to permit conclusive diagnosis in the individual case. Laboratory studies have to date proved negative. The similarity of symptoms to those of anxiety reaction and neurosis of various forms served to make the recognition of individual cases difficult. For these reasons, the positive diagnosis of cases must, we believe, be yet confined to the endemic situation.

Common factors between the separate reported epidemics include prolonged debility, symptoms of fatigue; pain in the head, neck, back and extremities; nausea and vomiting; and emotional disturbances of greater or lesser severity; a virtual absence of fever; a paucity of physical findings; and a lack of abnormal laboratory findings.

Epidemiologically, the confinement of cases to adolescent and adult patients and the significantly greater severity of cases among women are central to, and mandatory for the diagnosis.

It should be emphasized that the clinical picture presented in Punta Gorda will, in all probability, not present identically elsewhere. As each of the epidemics is reviewed in detail, it is apparent that there is a similar but not identical clinical and epidemiologic pattern of illness. In symptom detail and in over-all severity, there are clear differences.

Noteworthy, in contrast to the epidemic in Punta Gorda, was that which occurred in Tallahassee. Without question, this latter outbreak was more severe, the illness even more prolonged, and the sequelae more disabling.

Conclusion

The need for further investigation of subsequent epidemics is apparent. Care must be exercised to avoid overdiagnosis and the use of the illness as a "wastebasket" for wholly unrelated psychoneurotic problems. On the other hand, failure to diagnose and to recognize this illness in its epidemic form may be equally damaging.

The authors wish to express their appreciation to Drs. Roscoe S. Maxwell and Robert H. Shedd of Punta Gorda; to the staff of the Charlotte Hospital; to the volunteer citizens of Punta Gorda for their assistance in the house-to-house survey; to Dr. E. Charles Kunkle, Professor of Neurology, Duke University Hospital and Medical School; to Dr. Seymour Kalter and Miss Rachel Gorrie, Laboratory Branch, Communicable Disease Center; and to Drs. David Poskanzer and William Jackson Hall, Epidemiology Branch, Communicable Disease Center.

References

1. Bond, J. O.; Wolff, H. G., and Bistowish, J. M.: Unpublished data.
2. Jones, R. O.: Personal Communication.
3. Nayfield, C. L., and Hicklin, M. D.: Personal Communication.
4. Blattner, R. J.: Benign Myalgic Encephalomyelitis (Akureyri Disease, Iceland Disease) *J. Pediat.* 49:504-506 (Oct.) 1956.
5. Gilliam, A. G.: Epidemiological Study of Epidemic Diagnosed as Poliomyelitis, Occurring Among Personnel of Los Angeles County General Hospital During Summer of 1934, *Public Health Bulletin No. 240*, 1938.
6. White, D. N., and Burtch, R. B.: Iceland Disease, New Infection Simulating Acute Anterior Poliomyelitis, *Neurology* 4:506-516 (July) 1954.
7. A New Clinical Entity? *Lancet* 1:789-790 (May 26) 1956.

133 West Marion Avenue (Dr. Clement).