BORNHOLM DISEASE SURVEY
1956, 1957 and 1958

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Bornholm disease has been recognized for over a hundred years, and had been described by authors from many parts of the world, for example: Daae (1872) in Sweden, Finsen (1874) in Iceland, Dabney (1888) in the U.S.A., Williamson *et al.* (1924) and Pickles (1933) in England, Sylvest (1934) in Denmark, Scadding (1946) in Egypt, Hamburger and McNeil (1947) in India, and many others. In fact the disease appears to have no frontiers.

It was first brought into the limelight of world medicine by Sylvest (1930) a general practitioner from Copenhagen, who encountered the disease while on holiday with his family on the island of Bornholm in the Baltic. Dr William Pickles (1933) was the first to recognize it in this country from Sylvest's description, although the disease had been recognized under another name (epidemic pleurisy) by Williamson and others (1924).

After several attempts by many observers to find the cause of the disease, Curnen (1950) isolated the recently discovered Coxsackie virus (Daldorf and Sickles, 1948) from a sporadic case of Bornholm disease. Since then it has been repeatedly identified from faeces, throat washings, cerebrospinal fluid, by serology, and, for the first time, in human voluntary muscle by Lepine (1952) in France during the 1951 pandemic.

Present Enquiry

Method. In the summer of 1956 a very large outbreak of over 2,000 cases of Bornholm disease occurred in the town of Swansea, South Wales. Two hundred cases occurred in the recorder's practice of 9,500 patients, and he conducted a detailed clinical and epidemiological study of 74 cases which he had attended personally.*

As this was the first time for at least 35 years that such an epidemic

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had occurred in Swansea, it was thought important to study the extent of the outbreak outside the town, not only in South Wales, but in the British Isles. Information on the extent of the disease in South Wales was obtained through personal contacts with doctors in various localities, and the information thus obtained showed it to be confined to the south-west only. In the same year, Dr Swift of Winchester studied an epidemic in his own locality (Swift, 1957), and also organized a study of the distribution of other cases throughout the South-East England Faculty of the College of General Practitioners. At the end of 1956 the Epidemic Observation Unit of the College inserted a special notification form for Bornholm disease in the *Research Newsletter*. On this, members were asked to inform the unit whether or not they had seen any cases of Bornholm disease in 1956, and, if so, to state how many and at what time of the year these had occurred.

As a result of information received from various sources, a picture was obtained, retrospectively, of the extent of the disease in the British Isles in 1956. (Epidemic Observation Unit, 1957.)

Following this preliminary survey a further, more detailed, study was carried out in 1957 and 1958, with particular emphasis on the clinical aspect of the disease. For this purpose, a questionnaire was designed so that the information could be transferred to Hollerith punch cards, which proved to be of considerable help in the final analysis. Bornholm disease continued to be included on the voluntary notification form in each issue of the *Research Newsletter* of the College. When the recorder was informed of cases, the required number of questionnaire cards were dispatched to the notifying doctor. The total number of reports received in 1956, 1957 and 1958 by all these various methods of notification was 472. These consisted of 108 positive, and 364 negative returns. The total number of cases reported were 2,470, of which 2,000 were the estimated number in the Swansea epidemic. 186 questionnaires were satisfactorily completed, and the data from these provided the material for the clinical analysis. (Table I.)

I

CLINICAL ANALYSIS

Bornholm disease may be described as an acute, febrile, and highly infectious illness, characterized by the sudden onset of paroxysms of severe costodiaphragmatic pain and accompanied by an intermittent fever, anorexia, profuse sweating, and frontal headache. It usually runs a course of about six days ending abruptly with no ill effects, but it may last for more than a month.

An important characteristic of this disease is that the symptoms,

especially pain, show the phenomenon of remissions and exacerbations. Between episodes of pain there can be a period of complete recovery lasting from a few minutes to a few days. In spite of the severity of pain, the patients are usually not otherwise very ill and complications are rare.

The incidence of the various symptoms found in this survey are shown in table II, but they are described later in more detail.

TABLE I

NOTIFICATIONS OF BORNHOLM DISEASE.

Year	*Method of notification*	*Number positive reports*	*Number of cases*	*Nil returns*
1956	*Research Newsletter*	27	133	89
1956	S.E. England "Yellow Warning" ..	17	118	38
1956	Verbal reports from S. Wales ..	9	2,051	18
1957	*Research Newsletter*	28	60	63
1958	*J. Col. gen. Pract.*	28	108	166
Totals	.. ..	108	2,470	364

TABLE II

MAIN SYMPTOMS.

Symptoms	*Number of cases*	*Percentage of total*
Pain of muscular origin	186	100
Fever	160	86
Sweating	122	65
Headache	117	62·9
Anorexia	114	61
Nausea	87	46·8
Sore throat	50	24·7
Vomiting	40	21·5
Rigors	18	9·7
Photophobia	11	5·9
Diarrhoea	8	4·4
Pain on moving the eyes	6	3·1
" Giddiness " or " dizziness "	2	1·1
Frequency of micturition	2	1·1
Delirium	1	0·6
Total cases	186	

As an introduction to the clinical analysis, here is a verbatim

account of the disease, given by a sufferer who is a nurse at the recorder's practice. She has both intelligence and a sense of humour:

Thursday, July 26th was for me a perfectly normal day as far as my health was concerned, but by the early evening I was suffering, as I then thought, from a particularly nasty bout of indigestion. A few proprietary antacid tablets, however, relieved me of most of the discomfort and I returned to bed none the worse for eating so much cucumber. After a somewhat restless night I awoke about 4.00 a.m. suffering from a severe pain around my chest and back. The pain was much worse on breathing: I felt as though an iron band had been put around me and was being tightened at every breath I took. In fact, had I not been so fond of life, I might have stopped breathing altogether, to get rid of the pain, but I contented myself by taking as little air as possible into my lungs.

Not wishing to disturb the doctor, I suffered, but not in silence, for quite involuntarily I started emitting a series of grunts with each breath I took. I am sure it would have put a young primipara in the throes of labour to shame. By 8.00 a.m. the pain had become less severe and I sent for the doctor.

During the morning, I had rather a nasty frontal headache, but the pain in my chest was bearable, except when I moved or breathed deeply. I had no temperature at this stage, nor complained of anything other than a headache. On being examined, I was told that there was no pleural rub, no neck rigidity, and my glands were normal.

The following day found me much better so I decided to get up. I still had the headache, but the pain in my chest was negligible. However, by the time I came downstairs I felt as though what little grey matter I possessed had been taken away and replaced by a large amount of cotton wool.

This state of affairs continued until the evening of the following day (Sunday) when *suddenly* the pain returned in all its fury and I hurriedly got back into bed. My temperature this time shot up to 103°F. and I sweated profusely. My head was heavy, I felt nauseous, and sure that I had fallen foul of influenza as well as Bornholm disease! The pain again lessened within an hour or two, but the headache, sweating, and nausea persisted until the morning after.

During Monday my temperature was raised, but on Tuesday it was back to normal. I felt very little pain and my head was more clear. I remained in bed until Saturday, and, apart from some tachycardia and some general weakness, I seemed to have recovered. The following Tuesday I returned to work, but, for approximately 10 days after, I suffered from what seemed to me muscular pains across my back. I thought at times I could hear a grating sound when I moved suddenly or took a deep breath, but this was confined to the right side, although the actual pain had been felt on both sides.

Age and sex incidence. Of 186 patients in this series, 87 were male and 99 female, with ages ranging from 6 months to 72 years. The highest prevalence was amongst children of pre-school and school age, up to the age of 15, with a second peak in the 30-35 age group—probably parents of affected children. (Table III.)

In Swansea a graph drawn of the ages of the persons in the infected households followed very much the same pattern as that drawn for those who had suffered from the disease (figures 1 and 2). It did not follow the pattern of the age groups of the town as a whole (figure 3).

It was also found in Swansea that where children and adults were infected in the same house, it was a child who was first infected,

TABLE III

AGE INCIDENCE.

Age group	*Male*	*Female*	*Total*
Under 1 year ..	1	—	1
1 —	9	15	24
5 —	29	27	56
10 —	12	13	25
15 —	8	7	15
20 —	5	3	8
25 —	2	4	6
30 —	6	12	18
35 —	2	5	7
40 —	3	2	5
45 —	3	3	6
50 —	3	1	4
55 —	—	2	2
60 —	3	3	6
65 —	—	1	1
70 —	1	1	2
75 —	—	—	—
Totals	87	99	186

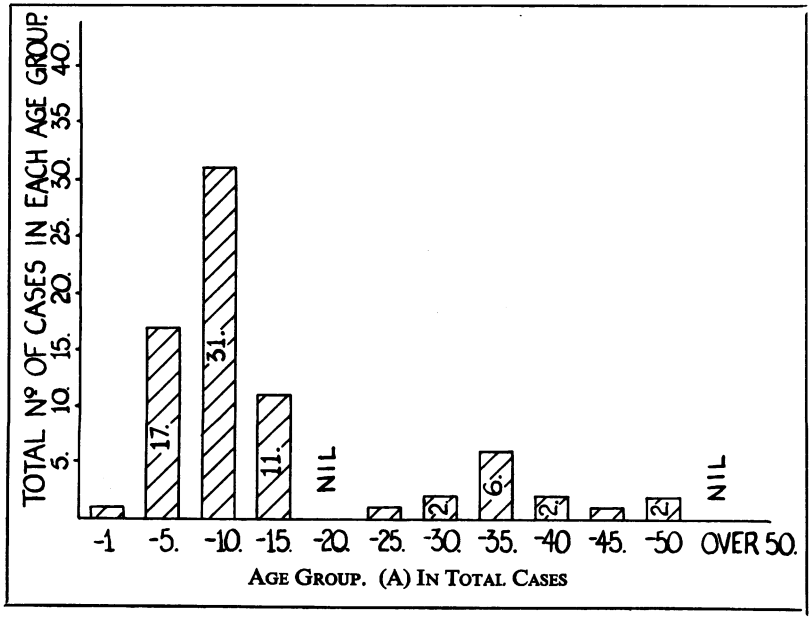


Figure 1

Age incidence of Bornholm disease in Swansea, 1956

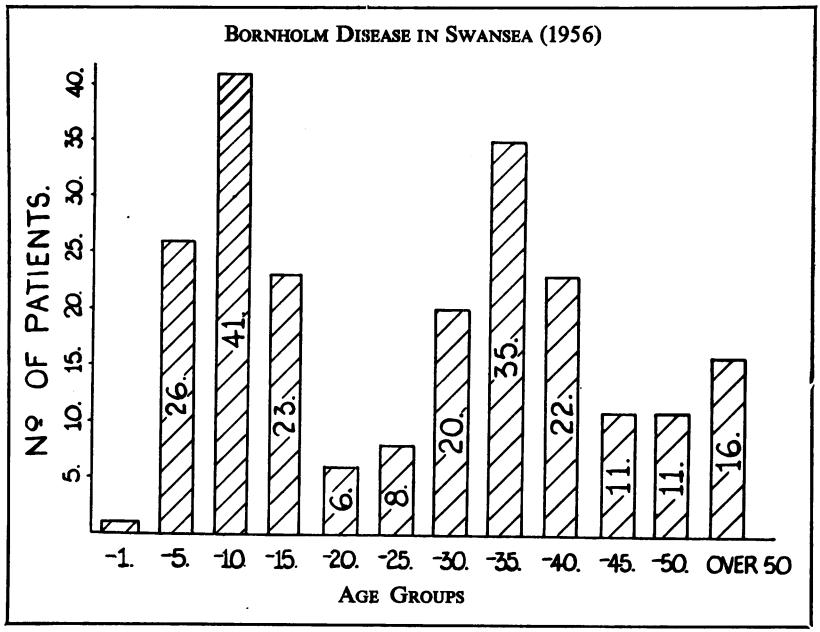


Figure 2
Number of patients at risk in each age group in the infected households

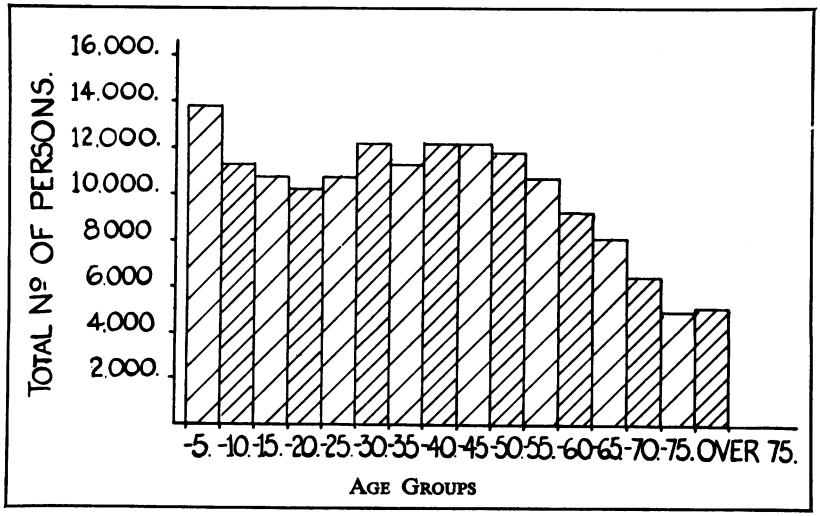


Figure 3
Estimated Population of Swansea Borough in 1956
(Calculated from 1951 Census Figures)

suggesting that the infection was picked up at school, then brought home to the parents.

Attack rate. The overall attack rate in the Swansea series expressed as percentage of cases amongst those at risk, was 34 per cent, this varying from 67 per cent in the 0—5 group to nil in the 15—20 group (table IV).

Of the rest of the persons at risk, the attack rates were highest amongst those of school age, and lowest in the 15—19 age group which had six persons at risk, none of whom developed the disease.

TABLE IV

ATTACK RATE OF BORNHOLM DISEASE IN SWANSEA, 1956

Age group	*Attack rate— as percentage of cases amongst those at risk*	*Age group*	*Attack rate— as percentage of cases amongst those at risk*
- 5	67	30	10
-10	75	35	17
-15	43	40	9
-20	0	45	9
-25	13	50	18
		over 50	16

Duration. The duration of the illness in this survey, varied from 1 to 40 days; in 53 per cent of the patients, the illness lasted less than 6 days (table V), and in only 2 per cent did it last more than 14 days. On the whole, the illness was of short duration, rarely lasting more than a fortnight.

The onset. The onset in many of these cases was sudden, often dramatic, causing a considerable amount of anxiety amongst the members of the household. This can well be imagined, as pain was by far the most alarming symptom.

Although the presenting symptoms in a disease may be dramatic or unpleasant to the patient, they may not necessarily mark the beginning of the illness. For this reason the " First symptom " was asked for on the College questionnaire. Here again, pain was in the forefront. Table VI compares the incidence of the first symptom with that of the presenting symptoms, and it shows that myalgia, especially that of costodiaphragmatic distribution, was prominent in most cases at the beginning of the illness, and even more so by the time the doctor was called in (presenting symptom).

Symptomatology

Myalgia. Each of the 186 patients suffered from pain at one time or another during the course of the illness. In the six-months old

TABLE V

DURATION OF ILLNESS.

Duration of illness (days)	*Number of patients*	*Percentage*
1	3	
2	6	
3	30	
4	32	
5	27	
		52·7
6	20	
7	12	
8	8	
9	6	
10	10	
11	11	
12	2	
14	12	
		43·5
17	1	
20	2	
21	1	
26	1	
28	1	
40	2	
		1·9
Not known	8	
		1·9
	Total 186	

TABLE VI

INCIDENCE OF (A) FIRST SYMPTOM AND (B) PRESENTING SYMPTOMS AMONG 186 CASES.

Symptoms	*Occurring as the FIRST symptom*		*Occurring as the PRESENTING symptom*	
	Number	*Percentage*	*Number*	*Percentage*
Costodiaphragmatic pain ..	130	69·9	160	86·0
Local muscle pain elsewhere ..	12	6·5	28	14·5
Frontal headache	2	1·1	25	13·5
General headache	2	1·1	30	19·4
Rhinorrhoea	2	1·1	5	2·7
Sore throat	5	2·7	24	12·6
Rigors	10	5·4	18	9·7
Malaise	11	5·9	39	20·9
Nausea	1	0·5	43	23·7
Vomiting	4	2·2	14	7·5
Diarrhoea	1	0·5	2	1·1
"Giddiness" or "dizziness" ..	1	0·5	2	1·1
"Fainted in the street" ..	1	0·5	0	0·0
Photophobia	0	0·0	2	1·1
Frequency of micturition ..	0	0·0	2	1·1
Constipation	0	0·0	1	0·5
Delirium	0	0·0	1	0·5
Pain on movement of the eyes	0	0·0	1	0·5
Any others	*1	1·1	†6	3·3
Not recorded	2	1·1	2	1·1

†Pain behind the eye and anorexia.
*Extraordinary weakness and difficulty in breathing.

infant this was judged clinically. The sudden and dramatic onset is illustrated from a case note:

> ... as I walked from the car to the house, I could hear a child's screams coming from within. The door of the house was open and a neighbour was waiting in the doorway. Inside there were more neighbours all looking agitated and worried. The mother was crying and holding the screaming child in her arms....

The pain can be so severe as to simulate pleurisy, coronary thrombosis, or an acute cholecystitis. Pain involving the muscles of respiration (diaphragm, intercostals, and accessory muscles of respiration) was most common, and occurred in 169 cases. In 50 of these the pain was referred to the abdomen, and in 20 to the shoulder tip. This can present a diagnostic difficulty because of other acute medical and surgical possibilities.

A characteristic aspect of this pain is that it is made worse on deep breathing, coughing, etc. In this series, 146 (78.5 per cent) of the patients showed this phenomenon. The recorder found that an increase in diaphragmatic activity made the pain worse, whilst holding the breath or clasping the chest eased it. The pain can wake a child from sleep, possibly due to an increased respiratory rate which in turn has been caused by an increase in temperature (Williams, 1959). This was found to be more pronounced in very young children.

Other muscles shown in this series to be involved were the limb muscles, psoas, trapezius, latissimus dorsi, and also—in six cases—the external ocular muscles of the eye.

Another important characteristic of Bornholm disease, confirmed in this survey was the recurrence of symptoms, especially of pain. It recurred in 118 (63.4 per cent) patients after an interval varying from a few hours in 85 (45.7 per cent) patients to a few days in 33 (17.7 per cent). In the remaining 69 sufferers pain did not recur.

Various authors have mentioned the shifting character of the pain (Dabney quoted by Sylvest, 1934; Hamburger and McNeil, 1947; and Warin *et al.* 1953). Although this was not specially asked for in the college questionnaire, it was recorded in the 74 Swansea cases in 1956. This phenomenon was demonstrated in 48.6 per cent of the patients and presented in three ways:

Localization of a more generalized pain
Appearance of pain in a new site
Total disappearance of pain from an original site, although persisting in another part of the anatomy.

Pain rarely moved from the diaphragm, but it often shifted to it from other muscles as though the virus seemed to have a particular

affinity for this site, or because this and the intercostal muscles are never at rest even during sleep. Pain on deep breathing, coughing, yawning, etc., was present in 146 (78 per cent) of the patients in the present series, and in 18 of those, this symptom recurred.

Sweating, fever, and rigors. Sweating in Bornholm disease is often profuse and of the degree found in influenza. Of 122 patients who suffered from sweating, in 60 it was profuse, while in the remaining 62 it was moderate.

Fever was complained of in 160 (86 per cent) patients, and the temperature was recorded in 149 of these.

Rigors occurred in only 18 (9.7 per cent) but it was interesting to find that it was a first symptom in ten of these.

Headache. Headache was reported to have occurred at some stage of the disease in 117 patients; it was absent in 66, and not recorded in 3. In 79 patients the distribution was frontal, and in 7 of these it was unilateral. The remaining 36 had a generalized headache. A tendency to recurrence of headache was observed in only 8 patients, 6 with a frontal and 2 with general headache (table VII).

TABLE VII

HEADACHE AS A GENERAL SYMPTOM.

	Present			*Recurrent*
Frontal ..	73	unilateral 6		1
		bilateral 67		5
General ..	36	.. ..		2
Absent ..	74			
Not known ..	3			
	186			8

Gastro-intestinal disturbance. Symptoms of gastro-intestinal disturbance occurred in the following frequency: anorexia 116 (62 per cent), nausea 87 (46.8 per cent), vomiting 40 (21.5 per cent), and diarrhoea 8 (4.4 per cent), (table VIII).

Among the Swansea cases anorexia was not severe and when the last episode of pain had disappeared, appetite was rapidly restored.

Vomiting, which was usually mild, did not occur without nausea. It was unusual for the patient to vomit more than once or twice, and diarrhoea, when it occurred, was very slight.

Table VIII shows that, although the virus is an enteric one, gastro-

intestinal symptoms did not have a tendency to appear at the beginning of the illness, but only after the disease had become established.

TABLE VIII

GASTRO-INTESTINAL SYMPTOMS: 186 CASES.

Symptoms	*Groups of patients with gastro-intestinal symptoms (see below)*		
	A	*B*	*C*
Nausea	87	1	43
Vomiting	40	4	14
Diarrhoea	8	1	2

A. Number of patients having the symptom at some time during the illness.
B. Number of patients where it occurred as a First symptom.
C. Number of patients where it occurred as a Presenting symptom.

Respiratory symptoms. Sore throat was present in 50 (27 per cent) patients; only in 5 of these did it occur as the first symptom. Unfortunately, information on the presence of a cough was not asked for in the college questionnaire, but among the 74 Swansea cases this occurred in 9 (12.2 per cent) of patients, although one of these already had a cough before the illness started. In Swansea also, the respiratory symptoms were much more in evidence at the beginning of the illness than were those of the gastro-intestinal system.

Photophobia and delirium. Eleven patients suffered from photophobia and one from delirium, but none had other physical signs suggesting a lymphocytic meningitis which sometimes complicates Bornholm disease.

Frequency of micturition. Frequency of micturition was reported by Walton (1951). It was present in three patients in this series, and was a presenting symptom in two. The third patient, however, was 3 months pregnant, and the symptom had been present before the onset of the disease.

The Effect of Age on Symptomatology

An analysis was made to find whether there was any significant difference in the incidence of the symptoms in different age groups. Among symptoms listed in table II, significant age differences

could only be demonstrated in the following:

Headache. *Frontal headache:* All ages together, 43 per cent exhibited the symptom. Variations by age were as follows:

Under 5, 40 per cent (a slight deficiency). All age groups from 5 to 34 years showed an excess above the mean value. For the 5 to 34 group as a whole, 51 per cent had this symptom. At ages 35 and over, only 15 per cent had frontal headache. Although these differences are statistically significant, it is difficult to give a clinical explanation for this observation.

General headache: Here again significant differences in the percentages in each age group were found, and can be summarized by saying that at ages 0—10 a smaller proportion had general headache than at ages over 10, and at ages 35 and over, this symptom is particularly prevalent.

Rigors. These occur extensively at ages 45 and over in which group they were reported by 29 per cent as compared with 8 per cent at ages under 15, and 11 per cent at ages 15 to 44.

Malaise. This occurred below average level (23 per cent) at ages under 15, and above average level at ages over 15, particularly in the 15—24 age group. It is a symptom which is difficult to assess in children.

Nausea. It was recorded in 58 per cent of patients under 15; in 39 per cent of those aged 15—44, and 38 per cent of those over 45. Here again, although significantly different, these percentages might have occurred in a control group without Bornholm disease.

Physical Signs

Helpful physical signs were few, and indeed in most cases it would have been impossible to make a diagnosis on physical signs alone.

Temperature and pulse. A raised temperature was recorded in 149 patients, and reported but not recorded in another eleven patients (table IX). In 24 patients, they were reported as having been afebrile throughout their illness.

TABLE IX

MAXIMUM TEMPERATURE.

Maximum temperature	*Number of cases*
Reported as afebrile	24
Under 103°F.	114
103°F. and over	35
Fever present but not recorded	11
Not known	2
Total	186

Swift (1956), whose cases are included in this series, found that each recurrence of pain was accompanied by an increase in temperature. Although information on this was not asked for in the

questionnaire, the recorder's series in Swansea followed this same tendency.

The pulse rate was generally found to be raised in proportion to the temprature.

Tenderness of painful muscles. Tenderness of painful muscles was present in 120 (65 per cent) patients.

Figure 4 shows a method of eliciting subcostal tenderness by placing the right hand beneath the costal margin and gently pressing backwards, stage i. The patient is then asked to exhale, stage ii, and the examining hand is then gently passed upwards behind the lower rib margin. At this stage the patient is asked to " take a deep breath ", stage iii, and tenderness if present is then elicited.

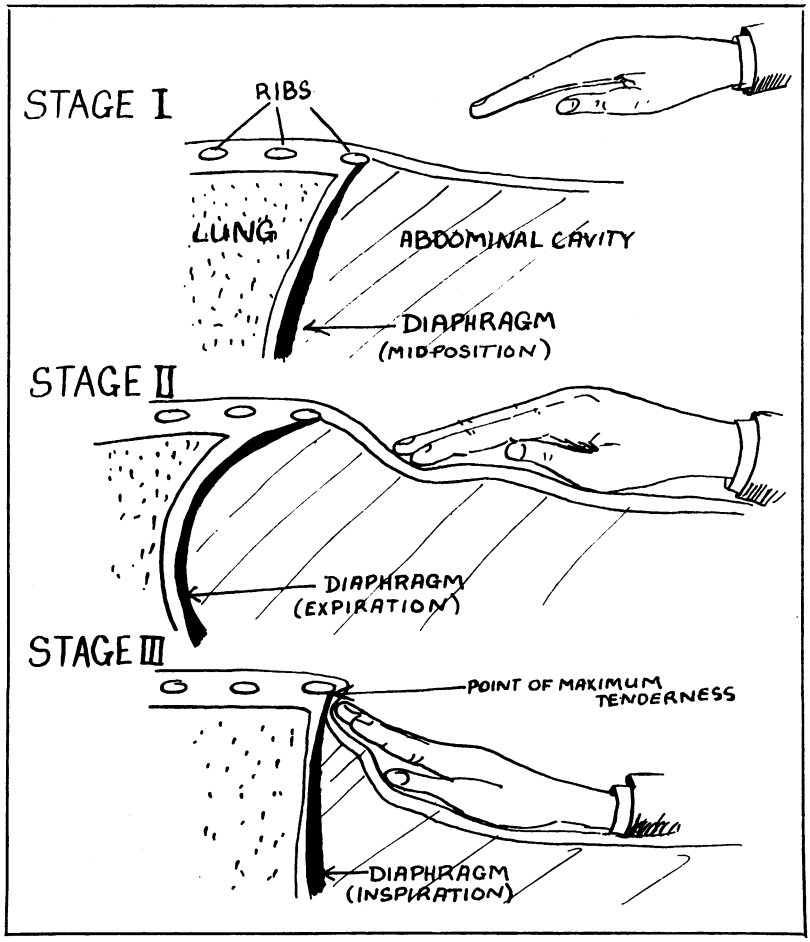


Figure 4
Method of eliciting subcostal tenderness in Bornholm Disease

The respiratory system. The respiratory rate in the whole series was recorded in 172 patients: in 106 the rate was normal; in 50 it was twenty to thirty per minute; and 16, over thirty. The highest recorded rate was 75 per minute in a child of 13 months, and the greatest increase in respiratory rate, as might be expected, was found in children under 5 years of age (table X).

TABLE X

RESPIRATORY RATES (MAXIMUM) AND GRUNTING RESPIRATIONS ACCORDING TO AGE.

Age groups	*Grunting respiration*	*Respirations (maximum)*				
		Normal rate	20—30	30+	*Not known*	*Total of patients*
Under 1 year	1	—	—	1	—	1
1— ..	9	5	6	12	1	24
5— ..	5	30	22	3	1	56
10— ..	2	19	5	—	1	25
15— ..	2	11	3	—	1	15
20— ..	1	5	2	—	1	8
25— ..	—	5	1	—	—	6
30— ..	3	9	6	—	3	18
35— ..	—	4	3	—	—	7
40— ..	1	2	2	—	1	5
45— ..	—	6	—	—	—	6
50— ..	1	3	—	—	1	4
55— ..	1	—	—	—	2	2
60— ..	1	5	—	—	1	6
65— ..	—	1	—	—	—	1
70—75 ..	—	1	—	—	1	2
Totals ..	27	106	50	16	14	186

An important characteristic of this disease was the frequency in which a grunting type of respiration was found. This occurred in 27 patients, with the highest incidence amongst children under 5 years of age (table X). There is no difficulty in recognizing the grunt, which occurs with each respiratory exertion. The sequence of events is as follows: (*a*) Inspiration brought to an end before it has reached its fullest extent; (*b*) a short pause or plateau; (*c*) immediately followed by a grunt marking the beginning of expiration.

Abnormal physical signs in the lungs in Bornholm disease are rarely found, unless the disease has been complicated by a pneumonitis or a pneumonia; a lobar pneumonia was recorded in one case. A " pleural " rub has been described in a proportion of outbreaks of this disease (Lloyd, 1924; Atlee *et al.* 1924; Scadding, 1946; Warin, 1953). In this series, seven cases were reported to have an audible friction rub. The cause of this type of rub is not quite clear, because it is different from the usual pleural rub, in that it is

loud, coarse, and extensive, and patients themselves can sometimes feel and hear it. The recorder is inclined to agree with Tidy (1951) who described the creak as " . . . loud and coarse, of a character which I have never heard in pleurisy, although in other instances it may be indistinguishable. . . . I agree with Sylvest in that it is not a pleural rub, but is of muscular origin, possibly from the sheath or tendon ". The friction rub might be compared with that found in tenosinovitis of the extensor tendon of the thumb. Another possibility is that it is a friction rub between the parietal pleura and the chest wall with its associated muscles. This would also explain the extensive area of the rub. The inflammatory processes could have spread from the muscles to the outer aspect of the parietal pleura.

Alimentary system. Stomatal ulcers were reported in two cases. Huebner (1953), in a study of 22 cases of epidemic pleurodynia in Texas, found ulcers in the throat in one case. He thought, however, that this might be herpangina because the only virus isolated in this particular patient was Coxsackie, group A, type 1, not group B as in Bornholm disease.

In the Swansea cases, some oedema of the palate and nasopharynx was found not only in patients who complained of a sore throat, but also in others in whom this symptom was absent. Forty-six out of 74 patients showed signs of inflammation in the palato-pharyngeal region, although only 16 of them actually admitted having a sore throat. The remainder, while not complaining of any discomfort, had definite oedema especially of the soft palate, with minimal reddening of the mucous membrane.

The findings on abdominal examinations were often negative; in others there were various degrees of tenderness, guarding, and rigidity, due to muscle pain. In some of the Swansea cases, the abdomen was board-like, quite as rigid as that found in peritoneal irritation following a perforated duodenal ulcer.

Four patients were admitted to hospital under observation as acute abdominal emergencies, and on one of these a laparotomy was performed, but nothing abnormal was found.

Neither liver nor splenic enlargement was observed in 180 patients; there was no record about 6 others.

Central nervous system. Neck rigidity, spinal rigidity, and Kernig's sign were reported absent in 180 cases (" not known " in remaining 6). One patient was reported to have had delirium and another had confused speech, but in neither were there abnormal findings in the central nervous system.

In some epidemics, lymphocytic meningitis has been reported

as having the same seasonal incidence as Bornholm disease, and may also occur as a complication. In Swansea in 1956, although it was estimated that over 2,000 cases of Bornholm disease had occurred in the town, not one case of lymphocyte meningitis was admitted to the local fever hospital during the period of the epidemic.

Conjunctivae. Bilateral reddening of the conjunctivae was recorded in 27 (15 per cent) patients, but none had unilateral reddening. All except one of these complained of sweating. In this survey the more severe the sweating, the higher was the incidence of conjunctival reddening; it was present in 30 per cent of those with profuse sweating but in only 13 per cent of those who sweated moderately.

Skin. Huebner (1953) reported seeing, on the third day in a child of 12 months, " a maculo-papular generalized rash suggestive of roseola infantum ". It should be noted, however, that he not only isolated Coxsackie virus, group B, type 3 from this case but also group A, type 1. A rash was reported in two cases in this series. One was described as having a blotchiness of the skin of the face, and the other as " a transient rash akin to German measles ".

Adenopathy. One treads on unsafe ground when describing the significance of palpable lymphatic glands in Bornholm disease. These are so common, especially in young children during the period of relatively rapid physiological growth of the lymphatic system. In 1956, the recorder found in the cervical region of several children who suffered from Bornholm disease strings of tiny glands about the size of apple seeds. When he examined a similar series of normal children afterwards, he found that this was common between the ages of 2 and 6 years.

TABLE XI

THE DISTRIBUTION OF ADENOPATHY IN THE PRESENT SERIES.

Adenopathy	*Number of cases*
Number with enlarged glands palpable	135
Tender cervical glands enlarged	9
Non-tender cervical glands enlarged	34
Axillary glands palpable	4
Inguinal glands palpable	4
Epitrochlear glands palpable	None
Not known	6

Relation to pregnancy

Three patients were respectively 12, 30, and 38 weeks pregnant when they developed the disease. Two had a normal confinement

but the third was complicated by a face presentation. The babies, however, were all normal. Facel Schaefer (quoted by Daldorf, 1955) reported two female cases who aborted in the 5th month of pregnancy following an attack of Bornholm disease. It is known that pregnant mice become progressively more susceptible to Coxsackie virus, group B as gestation advances and " many of the litter are lost " (Daldorf, 1955).

Complications

Complications which were reported in this series are listed in table XII.

TABLE XII

COMPLICATIONS

Complications	*Number of cases*
Mental depression	4
Right lobar pneumonia	1
Paroxysmal tachycardia	1
Right corneal ulcer	1
Sinusitis	1
Otitis media	1
Confusion of speech	1
Acute abdomen (observation)	3
Acute abdomen (laparotomy)	1
Orchitis	Nil
Lymphocytic meningitis	Nil
Abortion	Nil
Pericarditis	Nil
Encephalitis	Nil

In view of the relatively common occurrence of upper respiratory symptoms in this series, it was surprising that otitis media and sinusitis were not more common. One patient developed lobar pneumonia eight days after the onset of the illness. A doctor reported a child of 5 who, on the second day, developed a frontal headache, vomiting, confused speech, and "everything appeared big to him ". Mental depression was reported in four cases and lasted for as long as 3 weeks in one patient. Four patients, one of whom had a laparotomy, were admitted to hospital as acute abdominal emergencies.

The most interesting complication was found in a little girl aged 7 years who developed attacks of paroxysmal tachycardia on the seventh day of the illness and continued to have attacks for another 18 months. Coxsackie virus, group B, type 3 was isolated from the faeces of this patient. It is known that the Coxsackie virus, group B can cause a fulminating, often fatal myocarditis in new born infants (van Grevald, 1956; Verlinde *et al.*, 1956). The

supraventricular tachycardia in the 7 year old child may well have been precipitated by a mild myocarditis, resulting from a Coxsackie virus, group B, type 3 infection.

Certain complications described in the literature, but not observed in this series, have been included in table XII above.

Investigations

Faeces were examined in 51 patients and group B Coxsackie virus was isolated in 26 of these. Type 1 was isolated from 12 cases and type 3 from 14 cases. *Virological examination* was carried out with paired sera in 15 cases, but an increase in titre was found in only three, each infected with type 1 virus. *Throat washing* was performed in three cases but no virus was isolated. A leucocyte count was performed on 14 cases and found to be normal in all except one, where a mild leucocytosis was recorded. Out of 16 patients who had a *chest x-ray* there were only two with abnormal findings. One had evidence of an old traumatic haemothorax, and the other a " minimal evidence of pleurisy at left base ". *Laparotomy* was performed in one case but the abdomen was closed without any abnormality being found. Sylvest (1934) quoted a Danish surgeon as having attributed a case of peritonitis to Bornholm disease. At laparotomy, he had seen no abnormalities other than a mottled serous fluid.

Comparison between Patients with Type 1 and Type 3 Coxsackie Virus B Infection

There were 12 patients from whom the type 1 virus was isolated and 14 from whom type 3 was identified.

The main differences in clinical findings were found to be as follows:

Anorexia. Of those from whom Coxsackie group B, type 1 was isolated, 50 per cent suffered from anorexia, whilst in those infected with type 3, 93 per cent.

Pain referred to the abdomen. In the B1 group, 7 patients (58 per cent) had pain referred to the abdomen: in the B3 group none was recorded.

Respiratory rate. In B1 group, three patients (25 per cent) had a respiration rate above normal: in B3 group, ten patients (71 per cent) had a respiration rate above normal

Adenopathy. In the B1 group not one patient had a gland enlarged, whilst in the B3 group nine patients had enlarged glands.

These were the only clinical findings which showed a significant difference between the two types of group B Coxsackie virus infection. It would not be wise to draw any concrete conclusions from the above data, because the number of patients from whom the

virus was isolated was small.

Diagnosis and Differential Diagnosis

The diagnosis of Bornholm disease can be difficult, especially in sporadic cases. In an epidemic, however, once a typical case has been recognized, diagnosis of subsequent cases becomes easier.

A summary of the important diagnostic criteria is given below:

1. Usually abrupt onset
2. Fever with pulse rate in proportion
3. Pain of costodiaphragmatic distribution
4. Tendency for the pain to shift from one area to another
5. Symptoms, especially pain, show remissions and exacerbations
6. Pain made worse on breathing, laughing, coughing, etc., with absence of pulmonary signs
7. Grunting respirations in the absence of pulmonary signs
8. Loud, often extensive, friction rub in the chest
9. Frontal headache
10. Vomiting or diarrhoea (when present), very slight
11. Sweating, often profuse
12. Finding Coxsackie virus group B in throat washings and stools or evidence on serological examination confirms the diagnosis.

Difficulties may arise in differentiating Bornholm disease from other conditions.

(*a*) *Cholecystitis and cholelithiasis.* This difficulty was experienced, for example, in a 34 year old policeman whose pain had started in the right subcostal region, but which eventually moved to the left subcostal area. This made the diagnosis much easier, especially when he developed a frontal headache and a fever with profuse sweating. The appearance of the disease in other members of the household clinched the diagnosis.

(*b*) *Perforated peptic ulcer.* The difficulty in differentiating Bornholm disease from a perforated gastric or duodenal ulcer can be illustrated with the recorder's first encounter with this disease:

In the summer of 1951 he received an urgent call to see a young man, aged 19, who developed a sudden severe pain in the epigastrium. This pain was made worse on deep breathing, but there were no abnormal physical signs in the lungs. A provisional diagnosis of primary pleurisy was made, but later in the day he developed a pain in the right shoulder with an extension of the subcostal pain to the upper part of the right iliac fossa. There were still no abnormal physical signs in the chest, but there was some guarding of the muscles of the abdominal wall. Although there was no history of a duodenal ulcer, it was thought that there might be a leaking perforation of an acute peptic ulcer, with the gastric contents passing down the para colic gutter, and also upwards towards the diaphragm. He was admitted to hospital, but luckily escaped laparotomy because he recovered with amazing rapidity, and was discharged fit, three days later. Straight x-ray of chest and abdomen had proved negative.

His symptoms returned on the evening of his discharge, and he was promptly readmitted for further observation. He again recovered and further investigations failed to reveal evidence of an abdominal or thoracic catastrophe. The physician in charge was of the opinion that he was probably a case of Bornholm disease, and on looking back at the notes, there is no doubt that he was correct

in his diagnosis. The important criteria in this case were:

1. Frontal headache
2. Pain referred from the diaphragm to the right shoulder and made worse on deep breathing
3. No history of peptic ulcer
4. Absence of shock
5. No board-like rigidity (although this might not be present in a leaking perforation) but it does occasionally occur in Bornholm disease
6. Rapid recovery after the first seizure of pain followed by an even quicker recovery after the second and final seizure
7. No symptoms of an ulcer since the attack.

(*c*) *Acute appendicitis.* The degree of pain in acute appendicitis is usually much less severe than that in Bornholm disease, and, while pain settles down in the right iliac fossa in appendicitis, it usually flies to the subcostal region in the latter. Frontal headache, and indeed headache of any description, is uncommon in appendicitis (except when it has reached the stage of gangrene or perforation), but very common in Bornholm disease.

A furred tongue is common in appendictis but not in Bornholm disease. Rovsing's sign and tenderness on rectal examination are absent in this disease, but may be present as confirmatory signs in appendicitis. Psoas test was positive in one patient in this series, but this was thought to be due to the involvement of the muscle itself. The moral is wait and watch carefully, for patience will bring its reward. If, after waiting, there is still some doubt, it is wiser to admit the patient to hospital.

(*d*) *Pre-icteric stage of infective hepatitis.* There is often a marked nausea and vomiting, especially in children, in the pre-icteric stage of infective hepatitis; whereas in Bornholm disease both these symptoms are either absent or very slight. The stools and urine of Bornholm disease are of normal colour, and there is no abnormal pigmentation of the skin and mucous membranes.

(*e*) *Gastroenteritis and dysentery.* In these conditions, the diarrhoea and vomiting are usually severe, and the abdominal pain is more centrally situated. Bacteriological examination of the stool will usually give the answer.

(*f*) *Pyelitis, pyelolithiasis, and nephrolithiasis.* Absence of frequency of micturition or any abnormality of the urine help to exclude these conditions. Frequency of micturition has been observed in Bornholm disease in this series and also by Walton (1951), but, nevertheless, it is not a common symptom of the disease.

(*g*) *Pleurisy and pneumonia.* The typical creaking found in Bornholm disease has already been discussed. The rapidity with which the friction rub may cease or change location in Bornholm disease also serves to differentiate it from " dry pleurisy ". Indeed, such a diagnosis during the summer months might be thought of as a

missed case of Bornholm disease. An x-ray of chest and a white cell count is of considerable help in differential diagnosis; both are normal in Bornholm disease.

(*h*) *Prevesicular stage of herpes zoster.* In the recorder's experience herpes zoster is usually afebrile, and the pain is always located along the path of sensory nerves. Furthermore, in the prevesicular stage of herpes zoster there is usually no headache, sweating or pain on taking a deep breath.

(*i*) *Coronary thrombosis.* As in Bornholm disease, the pain of coronary thrombosis is usually severe, but seldom of such sudden onset, while the patient is usually shocked, with pallor of the skin, prostration and lowering of blood pressure. Taking a deep breath does not usually make the pain of coronary occlusion worse. One patient in this series, a man aged 48, had suffered from a typical attack of coronary thrombosis two years prior to the onset of Bornholm disease. He said in his own words " The pain was at times as bad as my heart attack ". An electrocardiogram showed no fresh myocardial lesion.

(*j*) *Influenza.* In both diseases, fever may start abruptly, and is accompanied by profuse sweating and frontal headache.

Photophobia is more common in influenza than in Bornholm disease, unless complicated by aseptic meningitis. The headache in epidemic myalgia is usually of shorter duration than that found in influenza. Aching in influenza is mild and generalized in contrast to the myalgia of Bornholm disease, which is severe and localized. The patients recover quickly in Bornholm disease, but in influenza convalescence is slow and the end of the disease is ill-defined.

(*k*) *Meningitis.* Symptoms and signs of meningeal irritation are absent in Bornholm disease unless complicated by aseptic meningitis, and it is in such cases that difficulties arise.

If there is any doubt as to the type of meningitis, a lumbar puncture must be performed without delay. In the meantime, specimens of stool, throat garglings, and serum should be examined for evidence of Coxsackie virus, group B infection.

Treatment

Method of treatment was not asked for in the questionnaire, but it was clear from the notes written on the back of the cards, from Swift's series, and the recorder's observations that no specific treatment was possible. The patients were given analgesics, usually in the form of aspirin, and also bed rest. In some epidemics the pain has been so severe that morphia has had to be administered. In the Swansea epidemic, aspirin in appropriate doses was given to

all the patients under observation in order to have uniformity of treatment. One should be very certain of the diagnosis before giving morphia, especially with cerebral or abdominal symptoms.

II

EPIDEMIOLOGY

Geographical distribution. The possibility of learning something of value from the geographical distribution of returns was investigated.

The size of each doctor's practice, involved in the survey, was obtained either by writing to him, or through the college records. The practice population represented in each county was compared with that of the county as a whole; this gave an idea of coverage.

The overall coverage for England and Wales, for example, was 14.7 per 1,000 population (England 14.8 and Wales 12.4). But the rate varied from " no returns " in several counties to 88 per 1,000 in one (Cardiganshire) where only one practice was represented. Not only England and Wales but Scotland, Eire and the Isle of Man, also showed a very low coverage rate.

With such incomplete coverage, statistics of the geographical distribution of the cases reported are of no value; therefore, these figures are not published.

Seasonal incidence. Epidemics of Bornholm disease, reported from all parts of the world, have occurred most commonly during the warmer seasons, whichever months they happen to be. Matheson (1947) for example, reported an epidemic in the South Pacific in 1947, which started on the Tongan Islands and about 20—40 per cent of the 16,000 inhabitants were involved; the Tongans and Europeans were affected equally. The low coral island of Tongatabu, although lying near the equator, experiences weather which is not exceedingly hot. The hottest time of the year is from January until March, and it was at that time that the epidemic occurred. Matheson also described another epidemic which occurred in the following year on Raratonga, the main island of the Cook group, which lies in the same latitude, but 600 miles east of Tongatabu. This epidemic also occurred during the hot season.

In the present survey there was a tendency for most of the cases to occur during the warm months of the year, June—October inclusive. (See table XIII and figure 5.)

TABLE XIII

DATE OF ONSET OF BORNHOLM DISEASE IN THE 186 CASES WHICH WERE STUDIED CLINICALLY.

Month of onset	1956	1957	1958	*Total*
January	—	1	—	1
February	—	—	5	5
March	—	1	3	4
April	—	6	1	7
May	—	3	2	5
June	35	4	5	44
July	36	4	13	53
August	1	4	10	15
September	11	2	1	14
October	30	1	1	32
November	—	—	3	3
December	1	1	1	3
Totals	114	27	45	186

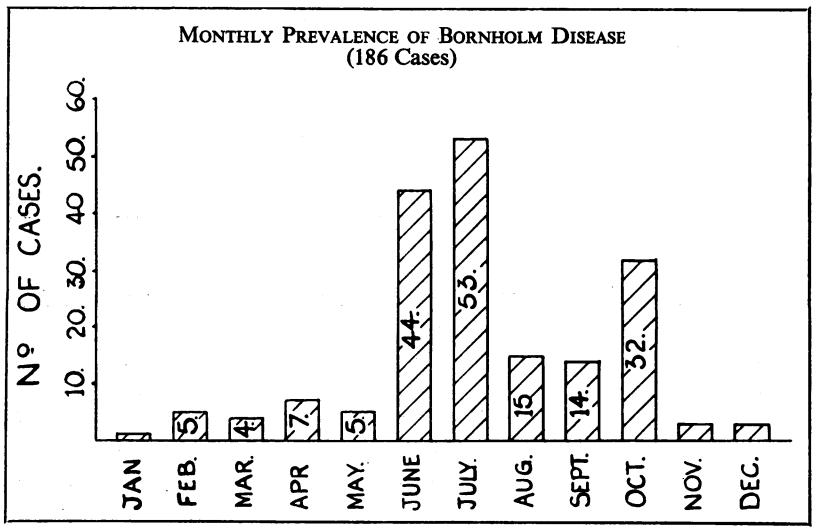


Figure 5

Though in line with poliomyelitis, " summer diarrhoea ", and other diseases due to infective agents living in the intestines, it is not known why Bornholm disease should be more prevalent in the warmer months, or why it should suddenly appear in epidemic proportions from time to time. In Swansea, for instance, the 1956 epidemic of Bornholm disease was the first in the living memory of the doctors who had practised for over 30 years in the town. In Winchester on the other hand, Swift mentioned a small outbreak which had occurred five years prior to the epidemic he studied in 1956. Bornholm disease does not usually spread like an influenza epidemic or a forest fire, it crops up here and there like mushrooms in a field, all growing together because the right conditions prevail. What these conditions are we are not certain.

The daily incidence of the 114 cases in the Swansea epidemic was interesting, because the proportion of adults to children stricken by the illness, increased considerably towards the latter third of the epidemic (see figure 6).

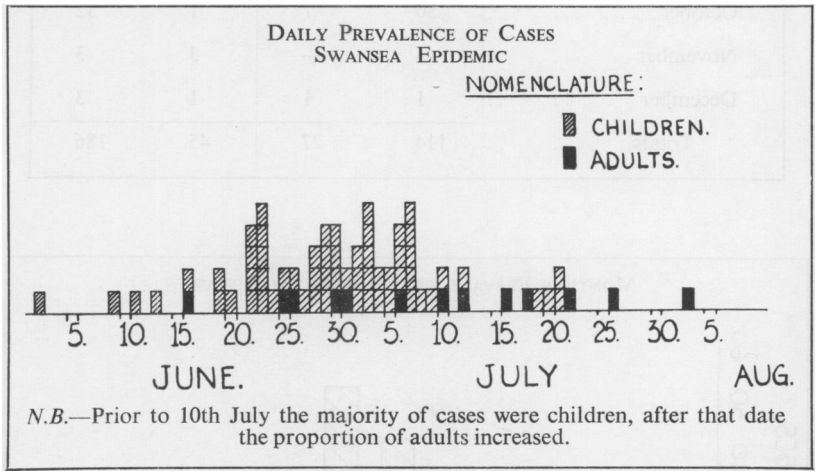


Figure 6

Duration of the illness. The duration of the illness was recorded for 178 patients. In 170 of these, the illness lasted from 1—14 days and in the remaining eight it appeared to have lasted a much longer period; in two cases as long as 40 days (see table V). In one patient the erythrocyte sedimentation rate was still raised after three weeks.

Place of exposure. Of the 186 patients studied, 61 were known to have been in contact with another recorded case. Fifty-four of these had been exposed to infection in the home, four at school and three in the houses of friends or relations. There was no

known contacts at work.

Incubation period. In this survey there were only 5 occasions on which a measurement of the incubation period could be made.

Child recently from America	3 days	
Child at birthday party	7 days	
1 " school child "	5 days	} See table XV
2 " elsewhere "	4 days	
	5 days	

TABLE XIV

SERIAL INTERVALS FOR CASE OF BORNHOLM DISEASE ACCORDING TO PLACE OF CONTACT.

Serial interval in days	*Place of contact*				*Total*
	Home	*School*	*Work*	*Elsewhere*	
0 ..	4				4
1 ..	3				3
2 ..	3				3
3 ..	9				9
4 ..	8			1	9
5 ..	3	1		1	5
6 ..	8				8
7 ..	6				6
8 ..	2				2
9 ..	2				2
10 ..	2				2
12 ..	1				1
14 ..	1				1
21 ..	1				1
25 ..	1				1
33 ..	1				1
Not known ..		3		1	4
Totals ..	54	4	Nil	3	61

Thus the incubation periods were 3, 4, 5, 5, and 7 days. Swift (1957) estimated the incubation period in his series to have been 4—7 days. Findlay and Howard (1950) infected a volunteer whose serum on numerous occasions had given a negative compliment fixation for Coxsackie viruses, by intranasal innoculation with a suspension of suckling mice in saline infected with Coxsackie virust B. Typical symptoms developed in 46 hours and in 118 hours complement fixing antibodies were detected in the serum. How this short experimental incubation period should be related to that following natural infection is not known.

For practical purposes the incubation period of Bornholm disease can be taken as 3—7 days, commonly 4 or 5 days.

Serial intervals. The serial intervals (Hope Simpson, 1954) in this survey varied from less than 24 hours to 33 days. A second case occurring in the same house in less than 3 days (the minimum

incubation period measured in this survey) probably represents a co-primary and not a true secondary case, but may mean the primary case was infective before the onset of pain and fever. There were 10 such cases out of 54 infected at home, but none among those infected outside the home. Thirty-four secondary cases infected at home, one at school and two " elsewhere " followed the primary case after the lapse of one normal incubation period—37/57 (65 per cent) of cases for which details are known.

Duration of infectivity. Eleven cases out of 54 (10 per cent) infected at home followed the primary cases by more than one normal incubation period, showing that the primary case was still excreting virus for several days after the onset of symptoms. The longest serial interval was 33 days, suggesting that excretion may have continued from this primary for not less than (33 – 7) 26 days the next longest period of infectivity was (25 – 7) 18 days which is in line with Swift's (1957) findings of positive isolations of Coxsackie B1 virus from three different patients on the 12th, 16th, and 18th days after the onset of their symptoms.

In the official report of the American Public Health Association, Bornholm disease (1955) it is stated that " the period of communicability is unknown, but apparently it is during the acute stage of the disease ".

Swift's observation (1957) that the virus can be isolated from the faeces for longer than 2 weeks after the onset of symptoms was confirmed in Swansea in two cases, when Coxsackie group B1 virus was isolated from the faeces 4 days after full clinical recovery of the patients. From further specimens collected 3 weeks after recovery virus was not isolated. Furthermore, clinical evidence of communicability of the disease after clinical recovery was shown in those two cases.

On 23 August 1957 (a year after the Swansea epidemic) a little girl celebrated her eighth birthday by entertaining a few friends to tea. Seven days later, on 30 August, she developed a severe pain in the chest and a grunting type of respiration. Her symptoms and signs were typical of Bornholm disease. Five days later, on 4 September, her young sister, aged 3 years 9 months, developed the disease and her symptoms were equally typical. Enquiries revealed that one of the guests at the birthday had been a boy of 3 years who had completely recovered from the disease a week before the party. The description given left no doubt that this boy also had suffered from Bornholm disease. None of the other children in the party suffered from such a disease either before or after the party.

It must be taken as established that a patient infected with Bornholm disease may excrete virus for at least a week, and possibly for 3 weeks, from the onset of symptoms.

Density of infection. The density of infection in each household was not asked for in the questionnaire, but the Swansea series

of 74 cases is shown in table XV.

TABLE XV

DENSITY OF INFECTION IN THE SWANSEA SERIES.

Number of houses	*Number of occupants per house*	*Number of houses showing number of cases of Bornholm disease per house*			
		1	2	3	5
12	3	12			
21	4	18		3	
13	5	7	2	4	
2	6	1	1		
2	7		1		1
1	9		1		
Total houses (51)		38	5	7	1
Total cases		38	10	21	5

Mode of transmission of infection. Evidence seems to point to transmission of infection by personal contact. Sylvest (1934) quotes an interesting paper by Green who observed an epidemic in the largest home for children in New York. Out of 1,230 children, 114 were affected. Extensive investigations had been carried out involving the personnel in the kitchen and the store rooms, also the persons who delivered food articles to the institution, but none of them seemed to have suffered from the disease. The milk was pasteurized by the institute's own farm. The water supply was not contaminated in any way. (Coxsackie virus had not been discovered as the cause of Bornholm disease until 1950.)

There were two separate departments in that school, one for the boys from 10—17 years of age, and the other for the girls aged 3—16. The two departments were facing each other, being separated only by a road, but during the epidemic the inhabitants were kept strictly isolated. The boys and girls were given the same milk and the same food, yet all the 114 cases occurred in the boys' section and none in the girls'. As a result of this investigation Green considered that the infection was spread by contact.

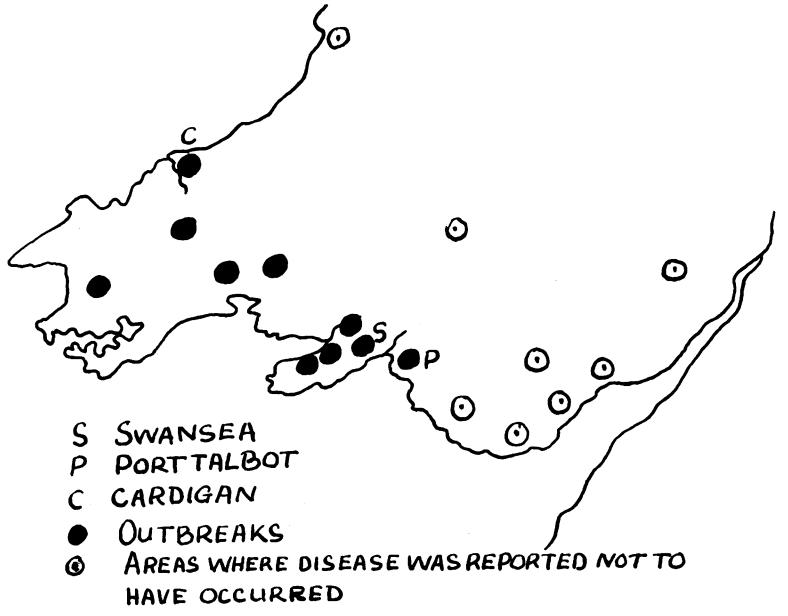
Daldorf (1955) reported that the Coxsackie group of viruses is more stable if suspended in dairy products, but that the standard methods for the pasteurization of milk had proved effective in destroying several of the Coxsackie viruses; but cream and to a lesser extent ice cream, give the virus more protection.

The possibility of transmission of infection through milk was considered by the recorder in the south-west Wales epidemic in 1956. It was difficult to explain why the epidemic should have been

confined to this area of south Wales (see map). In each infected household the housewife was asked where she obtained her milk and the children were asked if they drank milk at school. At the end of the epidemic a visit was made to the Milk Marketing Board headquarters at Llanelly in order to find out how the milk was distributed in south Wales. In one room there was a large wall map of south Wales with indicators of various colours to show:

1. Where the milk was produced (indicating individual farms) and where that milk was consumed.
2. The type of milk—whether pasteurized, sterilized, tuberculin tested, a combination of standards or not specified.
3. Where the milk was used for manufacturing purposes.
4. Clear areas where there were attested herds.
5. Designated areas where the milk has by law to be either tuberculin tested (raw), tuberculin tested pasteurized, pasteurized, or sterilized. (Swansea Borough is included in one of these areas).
6. To which parts of England and Wales the milk is transported in bulk.

Map of south Wales showing the distribution of outbreaks of Bornholm disease in 1956.



The following points of importance were learnt as a result of the information obtained from the Milk Marketing Board, and that obtained from the housewives from the infected households in the recorder's practice:

(*a*) Swansea Borough is a designated area.

(*b*) The town is supplied by a considerable number of retailers, some produce the milk themselves and others—the big dairies—receive it in bulk from farms, not only in the immediate neighbourhood, but from as far away as west Wales.
(*c*) The infected persons did not have a common milk supply in Swansea.
(*d*) East Wales (where no cases were recorded) also received milk in bulk from west Wales.

The recorder was unable to find evidence to prove that the epidemic in South Wales was spread by milk.

Most of the patients investigated in the college survey seemed to have been infected by another member of the household—someone in close contact, living, eating, and sleeping together. The exact mode of transmission could not be established. It may be by faecal contamination or by droplet infection. Possible spread by insects has not been investigated.

Comparison with other Virus Diseases in Swansea in 1956

(*a*) *Poliomyelitis.* There is experimental and epidemiological evidence that the poliomyelitis and the Coxsackie viruses interfere with one another. According to Daldorf (1955) it has now been repeatedly confirmed that several of the group B viruses modify the course of experimental poliomyelitis in mice. Figures for poliomyelitis in Swansea were obtained from the Medical Officer of Health. Table XVI gives the annual figures since 1947, and shows that there was no dramatic drop in incidents in 1956.

TABLE XVI

FIGURES FOR POLIOMYELITIS IN RESPECT OF NOTIFICATIONS RECEIVED FROM WITHIN THE SWANSEA BOROUGH FROM 1947 UNTIL 1957.

Year	*Paralytic*	*Non-paralytic*	*Total*
1947			31
1948	See	note below	11
1949			30
1950	24	24	48
1951	9	16	25
1952	4	12	16
1953	12	6	18
1954	3	4	7
1955	6	4	10
1956	6	6	12
1957	4	2	6

Note—Up to 1950 no separate figures were given for the paralytic and non-paralytic types. Inocculation against poliomyelitis began on 10 May, 1956.

Table XVII, on the other hand, shows that no cases of poliomyelitis occurred during the Bornholm epidemic in Swansea but there was one case in May, before it started, and six in the months of August and September after the epidemic was over. This may have been a coincidence, or the Coxsackie virus may have had an interfering effect on poliomyelitis virus infection during the months of June and July when the Bornholm epidemic was at its height.

TABLE XVII
KNOWN FIGURES FOR VIRUS DISEASES (SWANSEA 1956)

Month	*A*	*B*	*C*	*D*	*E*	*F*	*G*
	Bornholm disease	*Herpes zoster*	*Measles*	*Non-paralytic polio-myelitis*	*Paralytic polio-myelitis*	*Polio-myelitis (total)*	*Aseptic menin-gitis*
January ..	0	3	50	1	0	1	1
February ..	0	0	64	1	1	2	0
March ..	0	1	31	0	0	0	0
April ..	0	0	11	0	0	0	0
May.. ..	0	5	1	0	1	1	0
June.. ..	36	2	20	0	0	0	0
July ..	37	1	29	0	0	0	0
August ..	1	0	8	3	0	3	0
September ..	0	4	4	0	3	3	1
October ..	0	2	16	0	1	1	0
November ..	0	1	25	0	0	0	5
December ..	0	0	10	1	0	1	0
Totals ..	74	19	269	6	6	12	7

Note—(1) Figures for columns *A* and *B* were taken from the author's practice.
(2) Columns *C*, *D*, *E*, and *F* were the M.O.H's figures for the Borough of Swansea.
(3) Column *G*—figures for the Borough obtained from Hill House Infectious Disease Hospital.

(*b*) *Aseptic meningitis syndrome.* No cases presenting this syndrome were observed in the recorder's practice in the summer of 1956, but with the kind permission of Dr Pugh of Hill House Isolation Hospital, the case histories of all patients who had been admitted from the Swansea Borough suffering from meningitis in 1956 were read and all cases with aseptic (lymphocytic) meningitis were picked out. The results of these enquiries, given in table XVII, indicate that there were no cases of aseptic meningitis admitted to the hospital from the borough during the Bornholm disease epidemic. Dr Pugh was of the opinion that the five cases admitted in November were probably non-paralytic poliomyelitis, but they were not labelled as such because polio virus was not isolated. This suggests that Coxsackie B1 and B3 viruses do not commonly cause meningitis, though it is known that type B5 virus does.

(*c*) *Other virus diseases.* There was an abrupt increase in the preval-

ence of measles during the Bornholm disease epidemic, though this was probably a coincidence. Since 1954 the recorder had kept records of all cases of herpes zoster which he had seen in the practice. The prevelance of this disease was not significantly altered by the epidemic of Bornholm disease (see table XVII).

No cases of herpangina were recognized.

Summary and Conclusions

A clinical and epidemiological study of Bornholm disease carried out in 1956, 1957, and 1958 is described.

The disease was found to be most prevalent amongst those under 15 years of age, and the attack rate was also highest in this age group. Of the 186 cases studied clinically, there were 88 males and 99 females.

The illness usually started rather abruptly and lasted from 1—40 days. In 92 per cent of the patients its duration was 1—14 days, in 2 per cent 17—28 days and in two patients it lasted 40 days.

Myalgia, occurring in every patient, was the outstanding symptom, and was characteristically of costodiaphragmatic distribution in 86 per cent of the patients.

Other common symptoms included: fever 86 per cent, sweating 65 per cent, and headache 63 per cent. Gastro-intestinal symptoms (anorexia, 61 per cent, nausea 47 per cent, and vomiting 22 per cent) were more prominent after the disease was established, whilst upper respiratory symptoms were in evidence at the beginning.

In the Swasnea group of 74 cases, 46 had oedema of the soft palate with minimal reddening of the palato-pharyngeal mucosa, but it was found that only 16 of these actually complained of a sore throat.

Although the Coxsackie group B virus is an enteric virus, it is possible that it makes an entry into the body through the naso-pharynx and appears in the bowel later in the illness. The evidence in this paper shows that the virus may then be excreted in the faeces for at least a week, and possibly for 3 weeks after clinical recovery.

The respiratory rate was raised in 35 per cent of patients, the highest increase being in children under 5 years of age. A grunting type of respiration was a characteristic finding in the younger age groups, and occurred in 15 per cent of the group as a whole. Only seven cases were reported to have had a thoracic friction rub, the cause of which is discussed in this paper.

Another sign of interest was reddening of the conjunctivae, which occurred most commonly in those who suffered from a profuse

degree of sweating. This was probably due to simultaneous vasodilatation of the vessels of the skin and conjunctivae.

Although multiple small lymphatic glands were found in some of the younger children (0—5 years) this is thought to be due to normal physiological growth of the lymphatic system, as this phenomenon was also observed in a similar group of normal children.

Three patients who were pregnant when they contracted the disease, produced normal babies.

Complications were observed in only 14 patients—mental depression (4), right lobar pneumonia (1), paroxysmal tachycardia (1), right corneal ulcer (1), otitis media (1), confusion of speech (1), and 4 were admitted to hospital as acute abdominal emergencies, but on only one was a laparotomy performed—nothing abnormal was found.

Virus investigation carried out on a sample of 51 patients, showed 12 cases infected with Coxsackie virus B type 1, and 14 with type 3. A comparison of the symptomatology of these two types showed some significant differences, but no concrete conclusions were drawn.

The difficulties in differential diagnosis are discussed with several examples from actual case histories.

In Part II of the paper (Epidemiology) it is shown that because of insufficient coverage of returns from the whole of the British Isles an accurate picture of the distribution of the disease could not be given. The highest seasonal incidence in each year of the survey occurred in the warmer months, agreeing with that found in outbreaks in other parts of the world. The daily incidence of cases, as shown in the Swansea outbreak (1956), showed a greater proportion of adults affected at the end of the epidemic.

In the whole survey (186 patients), 61 (33 per cent) were known to have been in contact with other infected patients; 54 at home, 4 in school, 3 in houses of relatives and friends, but none at work. This supports the findings of Green that close personal contact is important in the spread of this infection.

The incubation period was found to be from 3—7 days and the serial interval varying from less than 24 hours to 33 days. Thus the duration of infectivity may be from before the onset of pain and fever for two to four weeks.

The density of infection in the Swansea group was shown to be sparse (1 per house) in the majority (38) of the 51 infected houses.

No evidence of spread of infection by milk was found in South Wales. Spread is possibly by droplet infection from infected persons in the early part of their illness and faecal contamination from those in the latter part of the illness and, later still, in the carrier stage. Close or prolonged contact at home favoured the spread of virus.

Interference of the polio virus by the Coxsackie virus is suggested as the possible cause of the absence of poliomyelitis in Swansea during Bornholm disease epidemic of 1956. Poliomyelitis occurred before and after the epidemic was over.

Although lymphocytic meningitis has been described as a complication and also occurring at the same time as Bornholm disease, no cases were reported in this survey, possibly due to the type of virus prevalent. No type B5 virus was isolated during the survey years from patients with Bornholm disease.

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