BORNHOLM DISEASE OR EPIDEMIC PLEURODYNIA.

A study of twenty-four cases and review of literature.

Introduction.

It falls to the lot of every general practitioner to see cases of illness on which he cannot put a proper diagnostic label, and which he designates as chills, or uses some other non-committal term.

The reason for this is that these conditions receive little attention in medical education, and are often not included, or are given very little space in the standard text-books of medicine.

One example of this class of illness is Exanthem Subitum or Roseola Infantum. This condition is much more common than is generally known and is often stated to be a chill with a sweat rash. The so-called second attack of Rubella is very often an attack of Exanthem Subitum. Another example is Glandular Fever, which, as Tidy (1951) has pointed out, is frequently overlooked.

Until lately the condition known as Bornholm Disease could have been included in this category of illness, but it is now becoming more generally known.

I cannot recall having heard the condition mentioned by any of my teachers when I was a student in the 1930s, and the two text-books of medicine

which I used viz. Price's Practice of Medicine - fourth edition, and Beaumont's Medicine - second edition, made no reference to it. This position has now been somewhat improved.

Most of the modern text-books make some reference to the disease, although the references are still rather brief and summary. The disease is much more common than they suggest.

Sylvest (1934) reported that from August 1930 to April 1934, 10,965 cases occurred in Denmark.

The association of the causal virus with the virus of poliomyelitis is attracting the attention of many physicians in various parts of the world.

In this country, the work of men like Pickles (1933-1939) and Hopkins (1950) has brought the condition before the eyes of the profession.

The hospital consultants are becoming more aware of its existence, as surgeons must keep it in mind when dealing with abdominal pain, especially upper abdominal pain in children.

At one time during the summer of 1951, a hospital in Fife had seven such cases in the surgical ward (Sandilands 1951). They had been sent in as abdominal emergencies.

During that same period, I had an outbreak in my practice in West Fife and I had the opportunity of

studying a series of cases. It is with this outbreak that I wish to deal in this thesis. I wish to draw attention to the manifestations of the disease and will base my description on notes taken at the time of the outbreak and when the cases were reviewed in the Autumns of 1951 and 1952. An abbreviated account has been given elsewhere (McNeish & Stewart 1951). Certain features of the condition will be described in detail e.g. the characteristics of the friction rubs, and certain conclusions will be drawn.

The findings will be compared and contrasted with those of other observers.

Historical Outline.

In this section, I propose to give a short historical outline of Bornholm disease as found in medical literature.

It will be seen that many names have been applied to the condition, but that on examining the reports, it is found that they all refer to the same disease.

In 1872 Daae described an epidemic of acute muscular rheumatism spread by contagion. This is considered by most writers to be the first published report on the disease.

The condition was seen by Finsen in Iceland in 1856 and 1862, but he did not publish his observations until 1874, and called it an epidemic of "stitch in the side" or "taksótts". Ronald (1942) states that the Swedish workers have shown that epidemics have occurred in the northern countries for centuries.

He points out that Waldschmidus described the illness in 1717, and that in 1735 Hannaeus gave a full account based on investigations in Schleswig, where it was known as Eyderstudishe Krankheit or "Stubble Fever".

Pickles (1939), quoting Prof. Major Greenwood, traced the disease in an outbreak in 1529 in the Haff of Stettin

- an outbreak which was part of the epidemiological phenomenom known as "English sweat".

As Daae's report is a fairly accurate description, I will quote it at length, as given by Akel (1944).

> "As a rule the patient has a stitch in one side "of the chest, most often without any precursory "ailment, but sometimes after an attack of chills, "the stitch is often accompanied by pains in the "back, neck, arms, and even in the fingers. "There is considerable difficulty in moving the "parts, especially the chest, therefore the "respiration is laborious, sometimes to such an "extent that the patient feels as if he were to "be strangled. Usually the general condition "is greatly affected. There is as a rule, some "headache, anorexia and thirst, and the bowels "are usually sluggish. The tongue is generally "coated. The pulse is normal or a little "frequent. There is seldom any cough, so cough "does not appear to go with this disease. "Physical examination of the chest reveals no "abnormality. There is a great difference in "the severity of the attack in various patients. "Some have a fairly mild attack and have to rest "only a short time. In others the attack is so "violent that one might expect them to die any "minute. In a few of the most severe cases, "the patients have got up and walked about as "soon as they have been able to do it. A great "many of them have had a relapse, sometimes "repeatedly. No case has terminated fatally. "Many of the patients are exhausted after the "disease, emaciated and feeble, sometimes they "feel a stitch or stabbing pain now and then for "several weeks after they have been able to begin "work".

The first account of the disease in American literature was given by Dabney in 1888. He observed an outbreak in Virginia and called it ? Dengue Fever. It was during this outbreak that the term "devil 's grip" was first used by a patient, a term which later on, was applied not infrequently in the United States.

In 1899 Reilly (cited by Harder 1936) described an epidemic of intercostal neuritis, which was probably the same condition.

Epidemics of the disease were reported in several states of America in 1923, 1924, and 1925. Various names were applied to these epidemics e.g. epidemic transient diaphragmatic spasm, epidemic pleurodynia, epidemic diaphragmatic pleurodynia. Epidemic pleurodynia is the one which has remained and is used at the present time in the United States of America.

In England, epidemics of "pleurisy" were described by Williamson, by Lloyd and by Attlee et alii in 1924. In these outbreaks, pleural friction sounds were noted.

It was not until the year 1930, that a full description of the disease, comparable to that given by Daae in 1872, was again given. This time it was a Danish physician named Sylvest who described the disease. He was on holiday on the island of Bornholm in the Baltic when he encountered 23 cases of what he called "Myositis Epidemica". In August 1930 he published a paper which gave the case records of his 23 patients. On the basis of the cases he had seen, he considered the disease was epidemic and that the most prominent symptom was pain in the abdominal muscles on inspiration. He presumed the condition to be a myositis localised especially to the upper abdominal muscles, the intercostal muscles, and other

muscles of the chest, besides, probably, the diaphragm. As he did not consider that the name "Myositis Epidemica" was suitable for laymen, he suggested that in Denmark at least, the disease should be called "den Bornholmske Syge" - Bornholm disease. This name was at once adopted by the whole Danish press and also by daily papers in other countries, especially when it turned out that, once the disease had been described, it could be found in many places.

In 1933 Sylvest published a further work in Danish on "Myalgia Epidemica or Den Bornholmske Syge", and in 1934 a somewhat briefer edition was published in English. In this monogram he reported on 93 cases and described tenderness and swelling of various groups of muscles and such complications as pleurisy and orchitis.

After the epidemic in Bornholm in 1930, Sylvest communicated with Pickles of Yorkshire and as early as 1933 Pickles was able to report cases which he had witnessed in his district. He was the first to use the name Bornholm disease in British Medical reports. Since 1934 Bornholm disease has been reported in many countries and in many climates. Outbreaks have been seen in Australia, New Zealand, U.S.A., the Tropics, Middle East and South Africa.

In 1946 Scadding reviewed the literature on the subject and reported on cases seen in the Suez Canal Hospital. Each year since then Bornholm disease or epidemic pleurodynia has been reported from various parts of the world.

Meningitis and meningo-encephalitis have been added to the list of possible complications. Gsell (1949) - cited by Thelin and Wirth (1951) has described cases of meningitis which he calls "meningitis myalgica".

Aetiology.

Bornholm disease has been known for some time to be due to an infection, but the actual site of infection and the pathology of the condition has not been fully elucidated.

In 1923 Payne & Armstrong reported an epidemic in Virginia U.S.A. and stated that the clinical signs pointed to involvement of the diaphragm or one of its serous surfaces. They called the disease "Transient Diaphragmatic Spasm".

Sylvest (1934), on the other hand, considered that groups of muscles were attacked and reported having observed tenderness and swelling of muscles in various places. He thought the diaphragm was sometimes involved, as several of his cases had hiccough, but that this was only part of a general muscle involvement.

This opinion was not shared by Locke & Farnworth (1936) who reverted to Payne & Armstrong's view that the disease was primarily an infection of the diaphragm. They pointed out that "the localisation of the pain "at the diaphragm level is constant, and that a very "important quality of the pain is its direct

"relationship to sneezing, coughing, laughing, deep "breathing, and especially exercise i.e. anything "that causes movement of the diaphragm".
Another exponent of Payne & Armstrong's view was Ronald (1942). He drew attention to the fact that the areas in which the pains are felt in Bornholm disease, are the areas in which Capps (1932) has shown that pain is felt when referred from the diaphragm. The diaphragm is innervated by the phrenic and the lower intercostal nerves, the former supplyimg the central, and the latter the peripheral part. Stimulation of the central portion gives rise to pain and stiffness in the neck through the phrenic nerve, and stimulation of the outer portions to motor and sensory reflexes referred to the areas supplied by the lower six intercostal nerves.

Ronald was of the opinion that the condition was either an infection in the diaphragm, or one which involved the diaphragmatic pleura. He pointed out that the diaphragmatic pleura is thin and is firmly connected to the underlying muscle, in contrast to the costal pleura which is tough and separated from the parietes by the endothelial fascia. Both pleura and muscle might thus be involved, but he also thought that the rapidity of resolution in Bornholm disease was possibly in favour of the lesion being muscular rather than pleural. He quoted Welborn (1936) as having

obtained a piece of latissimus dorsi muscle by biopsy from a patient suffering from Bornholm disease, and no morphological changes were found on examination. Ronald thought that if a piece of the diaphragm had been examined, some changes would have been found.

There would thus appear to be quite definite grounds for assuming that the diaphragm is the main site of attack, especially as will be shown later, the infecting agent is now known to have a predilection for muscle, although other tissues may be attacked as well.

Whether other muscles or muscle groups are involved in the inflammatory process is a point which will require further observation. Probably the correct view is that stated by Tidy (1951). He considered that the infection and pain are probably centred in the diaphragm and extend to the muscles above and below the costal margin. He reported that in a few cases he had seen definite swelling of muscles.

An entirely different view on the aetiology of Bornholm disease was expressed by Scadding (1946). He thought the condition to be a pleurisy since one in three of his cases had pleural friction and he named it "Acute benign dry pleurisy". This terminology had already been used by Williamson (1924)

Lloyd (1924) and Atlee et alii (1924).
Sylvester (1950) also considered the lesion to be a pleurisy, since he had X-Ray evidence of pleural oedema on the second day of illness, whereas Hopkins (1950) was of the opinion that when pleurisy did occur it was an extension from the diaphragm.

It is now clear that friction rubs have been reported in many outbreaks. Tidy (1951) considered that these friction rubs were muscular in origin, probably from the sheath or tendon.
The question of the causation of the friction rubs will be discussed in a later section of the thesis, when further evidence will be given suggesting that in most cases they are muscular in origin, but that true pleural friction rubs do occur although not so frequently.

The actual infecting agent has been a problem until recent years. Nasopharyngeal swabs from cases of Bornholm disease were examined by Greene (1924), but no specific organism was found. In the same year Small wrote a paper entitled "A Protozoan organism within the erythrocytes of patients suffering from epidemic pleurodynia". He gave microphotographs of the plasmodium and thought that he was able to differentiate it from the malarial plasmodium, but Sylvest (1934) made a search for the organism in a number of his cases and was unable to confirm its

presence.

During an outbreak of what was termed "Epidemic Pleurisy" in Eton College, England, Atlee et alii (1924) had throat swabs examined and both Streptococci and Staphylococci Aureas were found.

Streptococci were also reported in the nasal and throat swabs of patients examined by Cooper & Keller (1937) but no evidence was presented that these organisms had any relationship to Bornholm disease. The causal organism therefore remained unidentified for many years.

It was not until 1949 that the Coxsackie group of viruses was shown to be responsible by Curnen. This group of viruses was first described by Dalldorf & Sickles (1948). They published an account of an unidentified filterable agent isolated from the faeces of children with paralysis. The virus was named after the village of Coxsackie in New York state from which the patients came. Serological tests established that, after their illness, the blood of the two patients had antibodies capable of neutralizing the virus isolated in the faeces.

When working with other workers on a study of patients admitted to hospital suffering from poliomyelitis or aseptic meningitis, Curnen (1949) found one boy who had symptoms of Bornholm disease.

Examination of this boy's stools showed the Coxsackie virus and serological tests showed neutralizing antibodies in his blood.

At this time six of the laboratory workers who were working with the virus were infected. The Coxsackie virus was obtained from the stools and, or the throat swabs of each worker, and the capacity of the blood to neutralize the virus, which was not present before the illness, appeared early and increased to high titre during convalescence (Curnen 1950).

Other workers in this field of research were Findlay & Howard (1950) who were able to isolate a virus closely resembling Coxsackie virus from the blood, stools and nasal washings of a patient who had developed typical symptoms of Bornholm disease. Complement fixing antibodies for Coxsackie virus were present in the blood at the same time. One of them suffered a laboratory infection proved by a rise of specific antibody, the illness resembling Bornholm disease. They then obtained sera from 26 cases who had recently been infected in small epidemics of Bornholm disease. In all the cases the sera fixed complement in the presence of antigen made from Coxsackie virus strains. Negative results were obtained with sera from 26 healthy persons.

Strains of the Coxsackie virus were also isolated by Weller et alii (1950). They obtained the material

during an epidemic in the Boston area in 1947, but did not publish their findings until 1950. Specific neutralizing antibodies were also shown in the blood. Sylvester (1950) was another worker who reported positive complement - fixation tests to Coxsackie virus, in his series of cases, as did Ronse (1951) and Geffen (1951).
Geffen (1951) also reported isolation of the virus.

In 1952 Lazarus et alii and Brown et alii (1952) published reports of virus isolation and the demonstration of a rise in neutralizing antibody titre in typical cases of the disease.

It can therefore be categorically stated that the Coxsackie group of viruses is the infecting agent in Bornholm disease.

The characteristics of the group may be summarised as follows.
When injected into suckling mice, these viruses cause paralysis, probably due to lesions of muscle rather than of brain and cord. Their point of attack varies, however, as some strains attack brain, fat, pancreas or liver. They are of diverse serological types, placed in two broader groups A & B (Dalldorf 1950). Von Magnus at the International Congress of Medicine 1952, gave the number of types as 15.

Their seasonal prevalence is like that of poliomyelitis and in many instances they have been recovered from faeces along with the poliomyelitis viruses.

For this reason the Coxsackie virus and the Poliomyelitis virus have been referred to as fellow travellers.

Lazarus et alii (1952) have described the Coxsackie virus and the Poliomyelitis virus as being of the same order and size, and their physical properties as being closely alike. They also state that there is strong circumstantial evidence that the mode of transmission of both viruses is similar.

When further study and research have been done on this group of viruses, it may be possible to relate a certain strain of the Coxsackie virus to a certain form of Bornholm disease.

Epidemiology and Manifestations of the Outbreak.

The practice in which the outbreak of Bornholm disease was noted is centred in Crossgates, which lies between the towns of Cowdenbeath and Dunfermline. Cowdenbeath is two miles distant and Dunfermline is three miles.

The village of Hill of Beath lies one mile from Crossgates a little to the west of the Crossgates-Cowdenbeath road. Halbeath, another village, is also one mile from Crossgates and is on the Crossgates-Dunfermline Road. (Map No.1)

Both of these villages are served, as well as the outskirts of the two Burghs.

The practice can therefore be classified as semi-urban, semi-rural.

The combined population of the three villages is approximately 4000, and the inhabitants are employed mainly as follows:-

Crossgates - miners, industrial workers, dock-yard workers, agricultural workers.

Hill of Beath - miners.

Halbeath - industrial workers, miners.

My attention was first drawn to the

symptomatology of the illness in 1950, when Hopkins described cases which had occurred in his practice. It appeared to me that I had seen cases as he had described, but had not recognised them as Bornholm disease.

During the last week of May and the beginning of June 1951, cases began to appear in the district, but I was unable to put a label on the condition. When case No. 5 (see cases) came under my care, and I found one day that the pain and pleural rub were on the opposite side from where they had been when I last visited the patient, I realised I was not dealing with ordinary pleurisy. Then case No. 7 became ill. When first seen I thought this girl was going to have pneumonia, but the next day she was so well that I had to rule out this condition. Another child in the same household was now complaining, which meant I had two cases in the same house. This led me to think that I was dealing with something infectious or contagious, and epidemic pleurisy or myalgia suggested itself to me. On reading over Hopkin's article again, I became convinced that this was the same condition which was occurring in the area, and I collected notes on the individuals affected. Each case was reviewed in the Autumn of 1951 and again in the Autumn of 1952.

The notes on each case are given in detail below. The cases are recorded as they occurred.

Case No. 1 Boy age 8½ years.

This boy became ill late in the evening of 27th May. He complained of severe upper abdominal pain, and cried when any attempt was made to move him. He had no vomiting or diarrhoea and no headache. On examination his temperature was 99.4° F (37.4°C) and he appeared to be tender on both sides of the upper abdomen. On the left side the tenderness extended round to the renal angle, and on the right side it extended down to the iliac fossa. By the next day his pain was considerably less, and his temperature was normal, but the pain recurred the following day, and continued to recur until his parents sent in a call in the early hours of 1st June. When he was seen at that time, it was thought that he had a little nuchal rigidity with photophobia and a ? positive Kernig's sign, but later in the morning when seen again, the meningeal signs had passed off, and his pain was more localised to the lower left chest anteriorly. He was sent to hospital for investigation, and treated there as a suspected pneumonia. He was discharged on 11th June. A short time after he arrived home, he had a recurrence of his abdominal pain, which troubled him

intermittently for the next day or two, accompanied by a slight rise of temperature.

This patient was not recognised as a case of Bornholm disease, at the time, but in retrospect I consider that he was one.

There were no definite signs of pneumonia found before his admission to hospital, his illness had started with abdominal pain 4 - 5 days previously, and he had a recurrence of abdominal pain after discharge from hospital. His brother also had symptoms suggesting a similar condition and case No. 3, a typical case of Bornholm disease, was known to have been in contact with him. X-Ray of this boy's chest after discharge from hospital, showed no abnormality on the plate. When reviewed in Autumn 1951, his mother stated that he was having occasional attacks of pain in the right side of his abdomen. In February 1952 he was sent to Royal Hospital for Sick Children, Edinburgh, for investigation, as he was still having attacks of pain. He was admitted to hospital from the waiting list in June 1952. The case summary as supplied by the Royal Hospital for Sick Children, is as follows.

History - Admitted from the waiting list for investigation, with complaint of intermittent pain in the right rib margin, during the past year.

Examination -	No abnormalities found. Mid stream urine: culture - sterile.
Investigations -	Intravenous pyelogram : good bilateral kidney function. No abnormalities. No calculi.
Treatment -	12.6.52. Operation Appendicectomy with removal of Meckel's diverticulum.
Pathological Report -	The appendix shows no pathological changes. The mucosa of the Meckel's diverticulum is of the ileal type with abundant lymphoid tissue. There are no pathological changes.
Progress -	19.6.52. Discharged home. Need not report back.

When reviewed again in Autumn 1952, the mother stated that he had not complained of pain since his discharge from hospital. This is rather significant since no abnormality was reported at the operation.

Case No. 2. Boy age 3½ years.

This boy is a brother of case No. 1. He complained of vague abdominal pain on 26th May 1951, the day before his elder brother took ill.

The mother stated that it appeared to come in spasms, and that between the spasms he felt quite well. At the time the condition passed undiagnosed, the upset being attributed to some dietary indiscretion. His symptoms quickly subsided, but in retrospect, it is believed that he must have been subject to a mild attack of Bornholm disease.

When reviewed in Autumns of 1951 and 1952, he had had no further attacks.

Case No. 3. Male age 28 years. Miner,(local pit).

This man lives next door to cases Nos. 1 & 2. He was attacked by severe pain in the lower chest, on 4th June. In spite of the severity of his pains, he continued at his work underground. On 7th June he could not carry on any longer. The pain was now subcostal, especially on the right side. It was made worse by walking about, sneezing or even breathing. In the patients's own words, "the pains were like knives". He managed home with difficulty, but he could not lie down. When first seen he was sitting in a chair by the fire. He complained of intense pain on breathing. He also had a severe headache. No abdominal symptoms were complained of. He was sweating and shivering alternately . When examined, the abdomen was tense and guarded subcostally, and on deep palpation, he experienced pain at the right shoulder tip. The chest

was clinically clear. His temperature was elevated. Next day his temperature was normal and the pain less, but it continued to trouble him for two full days and nights. He was kept in bed for 10 days and allowed back to work in two weeks.

When reviewed in Autumn 1951 he reported no recurrence of pain. He was again reviewed in Autumn 1952. This time he stated he had had two slight attacks of pain lasting a few hours during the past year.

Case No. 4. Male, age 28 years. Miner, (non local pit)

This man was taken ill on the 4th June 1951. He was seized with severe pain in his back - lower thorax on both sides. The pain radiated round anteriorly and felt like a stitch. On 8th June, he reported to the surgery. By this time the pain was in the right hypochondrium and right lower chest, and was very severe on trying to take a deep breath, or on movement of the trunk. At the same time he complained of headache and a sore throat, together with a slight cough. There was no sickness, vomiting or constipation. Examination showed his throat to be injected. He was pyrexial. His chest was clinically clear. On examination of the abdomen, some tenderness in the right hypochondrium could be elicited. He was sent home to bed. By the next day his pain had almost gone.

His temperature had settled and his chest still revealed no abnormality. The pain gradually disappeared, and he was allowed up ten days later. Within a few days he was back to bed again on account of a recurrence of the pain. This time his symptoms were less severe and had entirely gone within five days. At no time were there any abnormal clinical signs in his chest. He had been feeling "run down" for some time before the onset of the illness, so he did not feel well for several weeks, but he finally returned to work on 23rd July, 1951.
Radiological examination of his chest showed no abnormality. There were no contacts of this case. When reviewed in Autumns of 1951 and 1952, he reported no recurrence of the illness.

Case No. 5. Female, age 35 years. House-wife.

On 12th June 1951, this patient reported at the surgery complaining of pain across the upper abdomen, which was worse on deep breathing. Her temperature was 99.8°F. (37.6°C) and her throat was injected. No abnormal clinical signs were detected in her chest or abdomen. Next day her temperature was normal, but she still complained of pain across the upper abdomen, which was catching her breath. By the 15th June, the pain was localised to the left lower chest anteriorly, and her temperature had risen to 102°F.

(38.8°C.). A friction rub was heard in this area on 16th June. A few days later the pain shifted to the lower right chest posteriorly, and a definite friction rub was present in this area also. This continued to be present until 29th June, when she appeared to be clear. Her temperature had been normal since 17th June, and except for a little pain on the right side posteriorly, she appeared to be quite well, but on 2nd July, pain in the left chest returned. Her temperature was again slightly elevated. Five days later she had completely settled except for a slight tachycardia, for which she was kept under observation for some time. This tachycardia gradually subsided. When reporting back towards the end of August, it was discovered that she had râles at the right base posteriorly. These had not been present in July. Radiological examination of her chest at this time was reported as showing no abnormality. There were no known contacts of this case.
When reviewed in the Autumns of 1951 and 1952, no recurrences were reported, and chest X-Ray plates again showed no abnormality.

Case No. 6. Male age 22 years. Labourer.

On 13th June 1951, this man had pain in the right subcostal region, shooting down into the right iliac fossa. The pain was worse on breathing. Two days

later he reported at the surgery thinking that he had appendicitis. There was no sickness or vomiting, and no elevation of temperature.

On examination there was localised tenderness over the lower part of right chest. Abdominal examination revealed no abnormality. Apart from the tenderness there were no abnormal clinical signs in his chest. Symptomatic treatment was given and he was feeling fit again in one week's time. There were no known contacts of the case.

When reviewed in the Autumns of 1951 and 1952, no recurrences were reported.

Case No. 7. Girl, age 10 years.

On 21st June 1951, this girl complained of abdominal pain at lunch time. When she returned to school in the afternoon, the pain got worse, and she was brought to the evening surgery.

She complained of severe pain across the upper abdomen and in the region of the right costal margin. The pain was excruciating whenever she took a deep breath. She also complained of headache. Her temperature was 101°F. (38.3°C.) and there was marked tenderness in the right hypochondrium. She had no sickness or vomiting. There were no definite abnormal clinical signs in the chest.

The next day her temperature was normal, she was

breathing quite freely and she had no pain. The tenderness in the right hypochondrium had gone. A day later she was feeling so well that she was allowed up, and in two days she was allowed out. This girl is a contact of case No. 8 and of cases 16 and 17 who live in the same block, and probably of case No. 9. When reviewed in the Autumns of 1951 and 1952, she reported no recurrence of the illness.

Case No. 8 Boy age 8 years.

This boy is a brother of case No. 7.
On 22nd June when I was visiting his sister, I was asked to see him, as he had a pain in his side. He complained of pain in the lower left chest anteriorly, which was catching his breath. There was a slight elevation of temperature to 99°F. (37°C). No abnormal clinical signs could be detected in his chest. He had no other complaints. The next day he was feeling quite well again. The pain had gone and he was allowed up.
No recurrences were reported when he was reviewed in the Autumns of 1951 and 1952.

Case No. 9. Female, age 28 years. House-wife.

This lady was seized with pain in the epigastrium and both hypochondria, on the afternoon of 22nd July 1951. In her own words "the pain was terrible". She also felt

shivery. There was no headache, vomiting, cough or irregularity of the bowels. She was seen by my assistant on 25th June and a tentative diagnosis of cholecystitis was made. When I saw her the next day the real significance of the symptoms was realised. The pain was then very much worse on breathing and seemed to catch her breath. Symptomatic treatment was given. She gradually improved within the next few days, but it was almost two weeks before the pain had entirely disappeared. No abnormal clinical signs were ever detected in the chest. This patient was a probable contact of cases No. 7 & 8.
No recurrences were reported when she was seen again in the Autumns of 1951 and 1952.

Case No. 10. Male, age 16 years. Miner (Local Pit.)

This boy felt a stabbing pain in his chest on 22nd June 1951, about 8 a.m. The pain was so bad he had to come home from work. His pain was worse on inspiration and he had it intermittently all day. Next day his pain had gone and it remained away until the evening of 24th June. He was suddenly seized with it again when sitting reading in bed. The pain was now across the upper abdomen and radiating to the lower left chest. Breathing was very painful, and he also complained of a severe headache. When seen about midnight, he had no cough or abdominal

symptoms. No abnormal clinical signs could be detected in his chest. He was tender on deep palpation in the left hypochondrium. His temperature was elevated. Next day his pain was less and his temperature normal. The pain gradually subsided and he was allowed up on 28th June. He reported to the surgery on 30th June, expecting to be allowed back to work, but on examination a friction rub was discovered at the left base anteriorly. This passed off a few days later, and he was signed off as fit for work one week later. Radiological examination of the chest revealed no abnormality. This lad lives a few doors from cases Nos. 1,2, and 3, but he was probably infected when at work.
When reviewed in the Autumns of 1951 and 1952 he reported no recurrence of the illness.

Case No. 11. Female, age 16 years. Factory Worker.

On Saturday 23rd June 1951, this patient complained of right-sided subcostal pain which was worse on breathing and was like a "stitch in her side". She was seen on Monday 25th June when her temperature was 100.2°F. (37.8°C.) and the pain was still very severe. She also had a sore throat. There was no vomiting or diarrhoea. Examination of her chest revealed no abnormal signs. Her throat was injected. Next day her temperature was normal. The pain lasted one week

and gradually disappeared, but recurred two weeks later for one day. This time the pain was very much less. Radiological examination of her chest revealed no abnormality.

When reviewed in Autumn 1951 she reported recurrence of pain two weeks after the initial attack, but in Autumn 1952, she had had no further recurrences.

Case No. 12. Male age 34 years Miner-(non local pit).

On 24th June 1951, this man awoke with a "stitch" in the left side of his chest. On the afternoon of the same day the pain returned when out walking. He could hardly get home. He went to bed but had to be propped up because of the pain which was now across the upper abdomen. He was seen on 25th June. His pain was now felt in the right lower chest anteriorly and was so severe that he said he was afraid to breathe. There was no headache and he had no abdominal symptoms. His temperature was 99°F. (37.2°C.). Examination of his chest showed no abnormality. Next day he felt much easier, but was still pyrexial, and on the following day he appeared to be back to normal. On 28th June he awoke again with a recurrence of pain, and recalled me to see him. This time the pain was epigastric and appeared to radiate more to the left lower chest. Chest examination again showed no abnormality and there was no elevation of temperature.

On 30th June when seen again, he had no pain, his temperature was normal and his chest was clinically clear. He was visited again on 3rd July. He was still apyrexial, had no pain, but had a definite friction rub at the right base anteriorly. This was still present on 5th July. By 9th July the rub was no longer heard, and the patient was feeling quite fit. Further examination at the surgery was negative as was also X-Ray report of his chest.

When reviewed in Autumn 1951 he reported no recurrence of the illness. He was seen again in Autumn 1952 - On 23rd September 1952, the patient presented himself at the surgery complaining of a recurrence of the pain which he had had last year. This time the pain was in the right subcostal region and along the upper border of the right trapezius. The pain had been coming in spasms and was much worse on breathing. Marked tenderness was present in the region of the pain in the subcostal region. A radiograph of his chest was again reported as showing no abnormality. This attack passed off in a few days and he was back at work on 30th September, 1952.

He stated that during the past year he had had three other similar attacks which had lasted two days, but they had been much less severe and had not kept him off work.

Case No.13. Male age 62 years. Miner (local pit).

This man was seized on the 27th June 1951, with severe epigastric pain "knife-like" in character and radiating to both sides of the upper abdomen. It was worse on deep inspiration and seemed to catch his breath. There was a slight elevation of temperature. He had no headache or abdominal symptoms. Symptomatic treatment was given. His pyrexia settled quickly and he was pain - free within two days and was able to start work on 2nd July. His chest was clinically clear throughout.

When reviewed in the Autumns of 1951 and 1952 no recurrences were reported.

Case No. 14. Male age 27 years. Miner (non local pit)

I was called to see this man in the evening of Sunday 1st July. He complained of pain in the right upper abdomen which was worse on breathing, and gave a history of a similar attack two days previously. The pain was very severe on deep breathing. He also complained of headache and had been given an aperient because of constipation. There was no vomiting. His temperature was 100°F. (37.6°C.). Marked tenderness was present in the right hypochondrium. No abnormal clinical signs were detected in his chest. He was seen next day and his temperature was now normal, his pain was less and he was feeling very much better.

Two days later the pain had completely gone. He was allowed back to work on 10th July. His chest at that time was clinically clear.
When reviewed in the Autumns of 1951 and 1952, no recurrences were reported.

Case No. 15. Male age 28 years. Miner (local pit).

On 30th June this man was seized with a sharp stabbing pain in the epigastrium and lower thorax, which was worse on deep breathing, coughing and sneezing. The pain became so severe that he sought medical advice on 1st July 1951. He had no vomiting or other abdominal symptoms. His temperature was elevated. No abnormal clinical signs could be detected in his chest or abdomen. The pain gradually subsided and he was back to normal on 5th July, and back at work on 7th July.
When reviewed in Autumns of 1951 and 1952 no recurrences were reported.

Case No. 16. Boy age 9 years.

On 1st July 1951, this boy had a pain across the upper abdomen. On 3rd July I was asked to see him. He had a temperature of 100°F. (37.6°C.) and still complained of upper abdominal pain. No clinical abnormality could be detected. On 6th July when seen again he was perfectly fit, his pain had gone and his

temperature was normal. He had probably been in contact with cases Nos. 7 & 8 who live in the same block of houses.
No recurrences were reported when his case was reviewed in the Autumns of 1951 and 1952.

Case No. 17. Male age 17 years Electrician.

On 4th July 1951, this lad was seized with acute pain in the right subcostal region, which was worse on breathing. He had no sickness or vomiting or other abdominal symptoms. His temperature was slightly elevated. On 6th July he complained of a stiff neck. Both sides of the neck were stiff. Symptomatic treatment was given. Next day the stiffness was much less marked and by the 11th July he was feeling quite fit again. He is a brother of case No. 16.
He was seen again in the Autumns of 1951 and 1952 and no recurrences of the illness were reported.

Case No. 18. Male age 16 years. Miner (local pit)

On 5th July this boy was seized with severe central epigastric pain which radiated upward into the left thorax. The pain was worse on deep breathing and coughing. He vomited once at the onset of the pain. There were no other abdominal symptoms and his temperature was normal. Examination of his chest and abdomen showed no abnormal signs except slight epigastric

tenderness. He was treated symptomatically. The condition quickly subsided and he was back at work in three days.

This lad was probably in contact with case No. 14, who lives a few doors from him and who had his attacks on 29th June and 1st July.

When he was reviewed in Autumns of 1951 and 1952, no recurrences were noted.

Case No. 19. Male age 20 years. Hairdresser.

This man reported to the surgery on 19th July 1951, complaining of having had a severe pain in the lower left chest, during the day. The pain was worse on breathing and on movement of the trunk. He also complained of a sore throat. He gave a history of having had a similar attack of pain on 16th July.

On examination he was running a temperature and no definite abnormality could be detected in his chest. He was sent home to bed. The pain and temperature settled in a very short time.

When reviewed in Autumn 1951 he reported that two weeks after his first attack he had a similar attack lasting for one night.

He was seen again in Autumn 1952. This time he reported that during the previous year he had had one slight attack of pain at the same site, for a few hours.

Case No. 20. Male age 17 years. Horse-driver.

On the morning of 19th July this lad wakened with bilateral subcostal pain. On rising he could not walk about because of the stabbing nature of the pain, which radiated upwards to both shoulders. He had felt shivery when at work the day before. He had no headache or sickness or other abdominal symptoms. On examination his temperature was 100° F. (37.6° C.) and he was sweating profusely. Breathing was inverse in type. The breath sounds were vesicular throughout both lung fields. Palpation of the upper abdomen was painful. His temperature was normal next day, but the pain was very severe for two days. He returned to work on 25th July, exactly seven days after the commencement of his illness. His sister, case No. 22, was affected on 24th July.

When reviewed in Autumn 1951, he gave a history of a similar attack of much less severity, two weeks after his initial attack.

He was seen again in Autumn 1952. This time he reported no recurrence.

Case No. 21. Male age 35 years. Joiner.

This patient attended at the surgery on 23rd July 1951, complaining of pain in the left lower chest anteriorly. The pain was very severe on breathing.

He gave a history of having had a sharp shooting kind of pain below both costal margins, on 13th July. The pain at that time made him "gasp" on breathing. From 13th to 20th July he had a constant "stitch" in his side which caught him whenever he walked or took a deep breath. The "stitch" was on the right side. By the 20th July it had passed off, but on the 23rd it had recurred, the only difference being that it was now on the left side. He stated that he had had a severe headache at the commencement of the illness and had vomited twice. When seen on 23rd July his temperature was elevated and he was sweating profusely. A friction rub was found at the site of the pain. This rub persisted for almost two weeks. Apart from signs of mitral stenosis which he was known to have, there were no other abnormal signs detected. By 10th August he was feeling very well, his temperature was normal, but a friction rub could now be detected at the left base posteriorly. This cleared up in a few day's time and he was back to work shortly afterwards. Radiological examination of his chest showed no abnormality except a mitral configuration of the heart shadow.

When reviewed in the Autumns of 1951 and 1952, no recurrences were reported.

Case No. 22. Female age 24 years. House-wife.

This patient was four months pregnant. On 24th July 1951 she sent in a call. She had been seized with acute right upper abdominal pain. The pain was so severe that she could not lie down in bed and had to be propped up on pillows. She was gasping for breath. At each attempt at inspiration she had severe pain. The pain she described as sharp, shooting, stabbing and knife-like in character and she had never experienced anything like it before. She also complained of a severe headache. Examination of the chest revealed no abnormality, but there was marked tenderness on the right side of the abdomen subcostally. Her temperature was normal. She was given morphia. Next day the pain was just as severe and there were still no abnormal clinical signs in the chest, but her temperature was elevated. On 26th July she was much easier, but she still had a temperature of 100°F. (37.6°C.). After a few days the pain and temperature gradually settled. When visited on 2nd August, a friction rub was detected at the right base anteriorly and she stated she could feel the rub when breathing. This was present for 10 days. She subsequently settled completely and an X-Ray plate of her chest showed no abnormality. She is a sister of Case No. 20.

When reviewed in the Autumns of 1951 and 1952 - no recurrence was reported.

Case No. 23. Female age 31 years. House-wife.

This patient became ill in the evening of 29th July, 1951. She complained of severe catching pain below both costal margins, and extending girdle fashion round the lower ribs to almost meet in the mid-line at the back. The pain was unbearable on inspiration and she had never had pain like this on any former occasion. She also complained of a headache, and pain up the back of the neck. There was no sickness or vomiting and no other abdominal symptoms were given. On examination she was seen to be sweating and highly fevered. Clinical examination revealed no abnormality. After a few days the pain subsided and the temperature settled. She continued to have slight attacks of pain after she was up and going about. Radiological examination of her chest showed no abnormality. When reviewed in the Autumns of 1951 and 1952, no recurrence was reported.

Case No. 24. Female age 54 years. Canteen worker.

This patient thought she had contracted influenza while on holiday on 18th July 1951. She did not feel too well for a week afterwards. On July 30th, 1951, she complained of sudden acute pain in the right lower

chest posteriorly. The pain was stabbing in character, especially on deep breathing and on walking. She also had a severe headache and sweated profusely. There were no abdominal symptoms or cough. Clinical examination of the chest revealed no abnormality. Her temperature was elevated. Next day her temperature had settled and the pain was less. In the course of a week the pain gradually subsided, and she was feeling fit enough to start work on 11th August. This patient was a frequent visitor to her son's home - case No. 21. When reviewed in the Autumns of 1951 and 1952, no recurrences were noted.

Having given the case histories I now wish to analyse the outbreak under different headings.

Location.

The map on page 41, shows the houses and districts affected. As can be seen the cases were distributed as follows:-

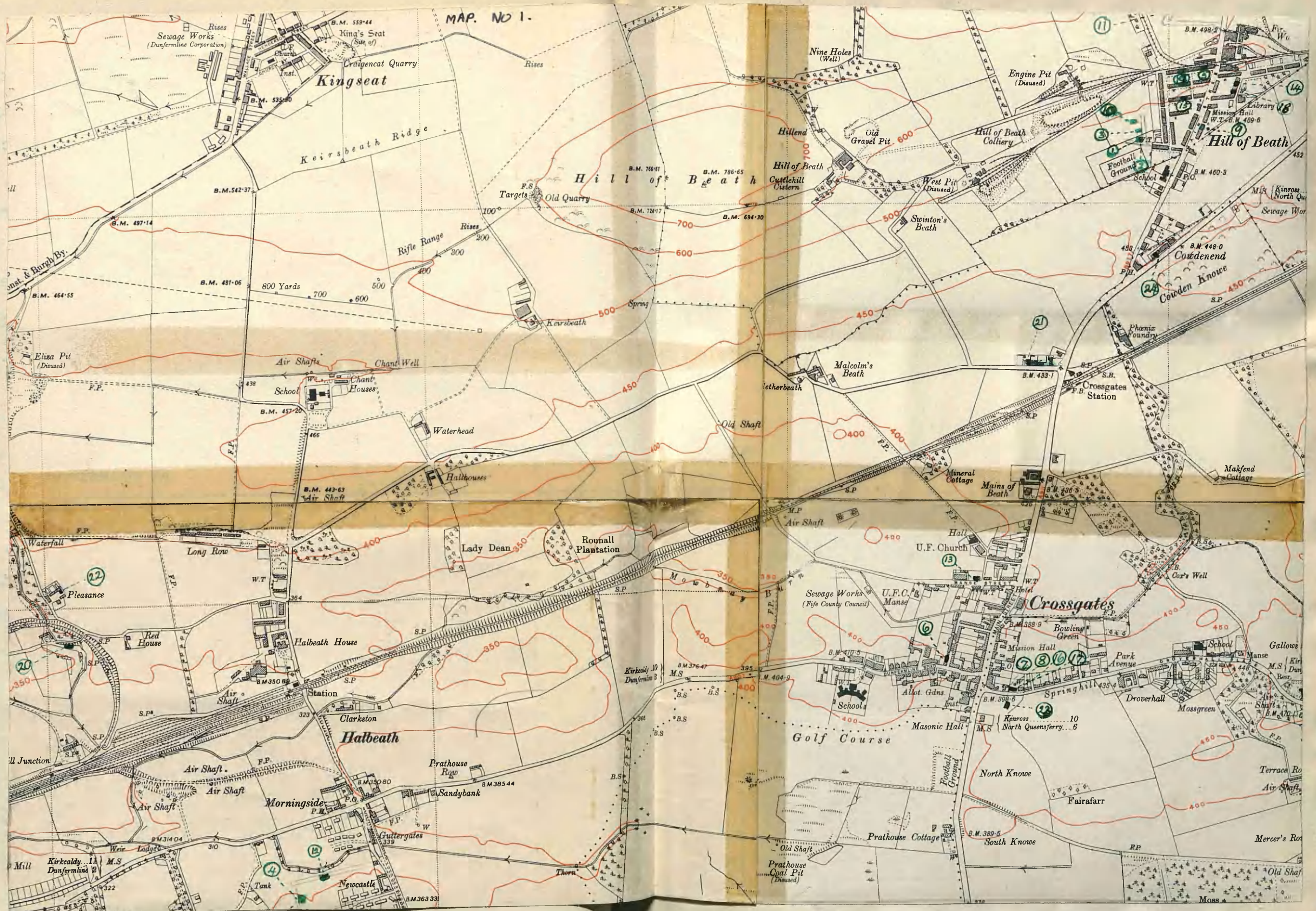
Crossgates	8
Hill of Beath	12
Halbeath	4

Since the centre of the practice is in Crossgates, the whole series will be referred to in later pages, as the Crossgates series.

MAP OF CROSSGATES AND DISTRICT.

MAP No.1.

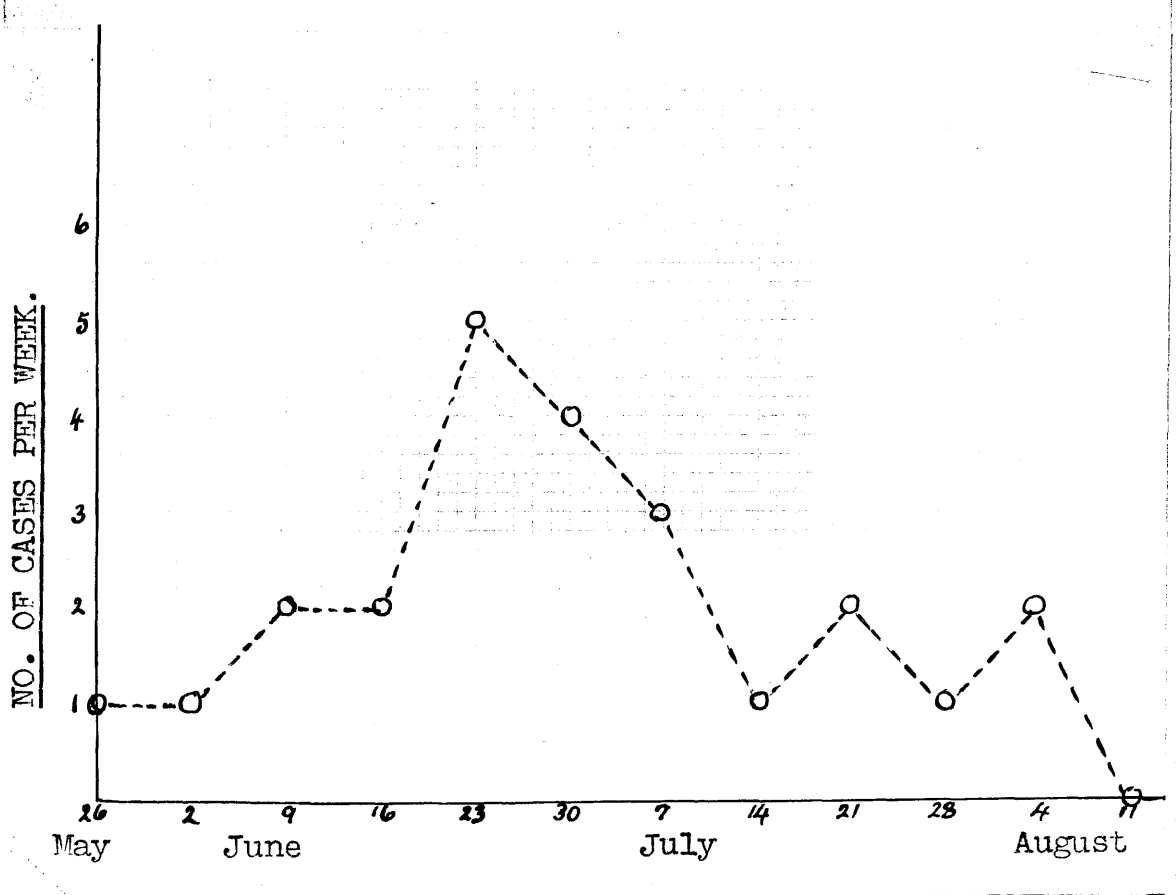
MAP. NO 1.



MAP No.1.

Incidence.

The first cases were seen at the end of May 1951, and the last one on 30th July, 1951.
The outbreak was therefore almost exclusively confined to the months of June and July.
A graph of the cases is shown below.
The peak period can be seen to be during the week ending on the 23rd June, when there was a sudden rise in the number of new cases, and then a gradual fall.



Graph showing new cases each week during outbreak.

Weather.

In Scotland during the month of May 1951, the weather was mainly cool, but dry and sunny. In June the dry sunny weather continued for 10 days, then it rained practically every day until the 20th, after which it was showery until the end of the month. The month was cooler than usual in most districts. During July the temperature maintained a moderately high level. The month was duller than usual, and on the whole, rainfall showed a considerable excess due largely to thundery downpours. (Registrar - General of Scotland). The weather would thus appear to have been cool, dry and sunny at the beginning of the outbreak, and warm, showery and dull at the end, with a period of heavy rain just before the peak period.

Infectivity.

Family Contact.

5 families had 2 members of the family affected

Nos. 1 & 2
7 & 8
16 & 17
20 & 22
21 & 24

It was noted that although 2 of the house-holds had large families of young children, yet only two members contracted the illness.

House to house contact.

As can be seen from Map No. 1, there was evidence of house-to-house contact having taken place.

No. 3 lived next door to Nos. 1 & 2.

Nos. 16 & 17 lived next door to Nos. 7 & 8.

No. 14 lived a few doors from No. 18.

No. 9's mother, whom she visited frequently lived below Nos. 7 & 8.

Contact at work.

Five of the cases were school children - 2 at Hill of Beath school, and 3 at Crossgates. There was no evidence of contact at school.

Miners.

Eight miners were affected; 5 were working in the local pit. The dates on which they first complained were as shown in Table No. 1.

As can be seen Nos. 10, 13, 15, and 18 occurred fairly soon after one another. Case No. 19 is a hairdresser and he was probably infected at his work, as he attended some of the miners who contracted the disease. It will thus be seen that family contact, house-to-house contact and contact at work were all responsible for the spread of the disease.

Incubation Period.

Owing to the difficulty in establishing definite times of contact, it was not possible to arrive at an exact incubation period, but from a study of a few

Case No.	Onset of Symptoms.
3	4th June
10	22nd June
13	27th June
15	30th June
18	5th July

Table No. 1

Table showing dates of onset of symptoms in miners employed in local pit.

Case No.	Onset of Symptoms.
Family { 16	1st July
{ 17	4th July
" { 20	19th July
{ 22	24th July
" { 21	13th July
{ 24	30th July

Table No.2.

Table showing dates of onset of symptoms in families where members affected were not infected from the same source.

of the cases, it appeared to be in the region of 3 - 6 days. As stated there were five families where two members were affected. In two of these families each patient appeared to have been infected from the same source and developed symptoms fairly soon after one another, (Nos. 1 & 2, 7 & 8). The dates on which the other families showed symptoms are shown in Table No. 2, and it is probable that in at least two of these families, one member infected the other. No. 14 possibly infected No. 18 and the dates on which they first complained were 29th June and 5th July. From a study of Table No. 2 and the above dates, it will be seen that 3 - 6 days can be taken as a probable time of incubation.

Age.

The ages and sex affected are set out in Table No. 3. Only five of the number were of school or pre-school age. This represents a little over 20%. Seventy per cent were under 30 years of age.

Sex.

Seventeen of the patients were males and seven were females. The miners who were probably infected at work may have caused the difference between the sexes.

Prodromata.

Only two cases (No.4 & No.24) complained of feeling "run down" before the onset of the illness.

Onset.

In nearly every case the onset was described as sudden, but almost half of the number did not seek medical advice until 1 - 12 days after the first symptom, and two of the series were only seen because I happened to be visiting the house at the time.

Symptoms & Signs.

Pain.

The outstanding symptom was pain in the upper abdomen or lower thorax. The site of pain and frequency of occurrence are shown in Table No. 4. As can be seen from Table No. 4, abdominal pain was met with in 20 cases (83%), and the thoracic type in 4 cases (17%). Of the patients with abdominal pain, seven had it in the right upper subcostal region. In the five children in the series the pain was mostly upper abdominal. Some of the patients who initially had pain in the upper abdomen, had pain in the lower thorax, on one or other side, within a few days. Others had transient pain in the lower thorax before the onset of the acute crippling pain in the abdomen. The pain was definitely related to respiration in most

No. affected	Age in years	Male	Female
4	Under 10	4	
5	Under 15	4	1
6	10 - 19	4	2
7	20 - 29	5	2
5	30 - 39	3	2
2	40 -	1	1

Table No. 3.

Table showing age and sex distribution of the cases.

Site of Pain	Frequency.
Abdominal	20
Right upper subcostal	7
Bilateral subcostal	3
Vague Upper	4
Central Epigastric radiating to either side	3
Epigastric spreading to lower left side of chest	3
Thoracic	4
Left lower anterior	2
Bilateral lower posterior	1
Right lower posterior	1

Table No. 4.

Table showing site and frequency of occurrence of pain

of the cases. In 20 it was much worse on deep breathing. Some complained that the pain was like a stitch in the side, others gasped on account of its severity and were afraid to breathe. Movement of the trunk and walking increased the pain in several cases. Various adjectives were used to describe the pain - terrible, knife-like, sharp, shooting, unbearable. The pain was intermittent and spasmodic in character in most instances and on account of this six of the male patients were able to continue at work for periods ranging from 2 - 10 days, when they were forced to seek medical advice because of the return of severe pain which had apparently left them for a day or two.

Relapses.

This relapsing character of the illness was an outstanding feature in cases 1, 5, 10, 12, 19, and 21. Each relapse was preceded by a period when the patient was apparently free from symptoms. The period lasted from 1 - 3 days. In cases 5, 12 and 21, the pain shifted from one side to the other side when the relapse occurred.

Referred Pain.

Patient No. 6 had pain radiating into the right iliac fossa, which made him think he had appendicitis. Another patient, case No. 17, complained of pain in the back of the neck as well as subcostal pain. Case No.23

also had neck pain, and pain in both shoulders was complained of by case No. 20.

Gastro-Intestinal Symptoms.

Sickness and vomiting at the commencement of the illness were reported by two patients (nos. 18 & 21) and only one complained of constipation (No.14.).

Headache.

Nine of the patients complained of headache at the commencement of the illness, five describing it as severe.

Fauces.

Four patients complained of a sore throat. In each case the throat was injected. These were Nos. 4, 5, 11 and 19.

Temperature.

All but three patients had a rise of temperature, ranging from 99°F. - 102°F. (37.2°C - 38.9°C). The temperature appeared to fall as the pain subsided and in two of the cases where there was a recurrence of pain, a corresponding rise in temperature was recorded, (cases No. 1 and 5).

Sweating.

Sweating was noted as a definite feature in five of the series, (Nos. 3, 20, 21, 23, and 24).

Abdominal Examination.

Upper abdominal tenderness was present in nine cases, being in the right subcostal region in five. One patient had pain in the right shoulder-tip on deep palpation in the right hypochondrium (case No. 3). Guarding of the muscles in the upper abdomen was also present in this case. No other abnormalities were noted on abdominal examination.

Chest Examination.

Examination of the chest at the onset of the illness revealed no abnormality except restricted breathing and occasionally inverse breathing. Tenderness over the lower right thorax was noted in one case (No.6). In five cases friction rubs were noted 4 - 10 days after the onset of the symptoms, as shown in Table No. 5.

The relationship of the pain and the friction rubs is shown in Table No. 6. As can be seen, three of the patients had no pain when the rub appeared. The character of the friction rub in case No. 22 was noted as being very coarse, and the patient stated that she could actually feel the rub.

Case No,	Age in years	Sex	Situation	Time from onset of symptoms	Duration
5	35	F	Left base anteriorly Right base posteriorly Recurrence to left base anteriorly	4 days 7 days 20 days	2-3days 10 days 5 days
10	16	M	Left base anteriorly	8 days	Less than 7 days
12	34	M	Right base anteriorly	9 days	3-6 days
21	35	M	Left base anteriorly Left base posteriorly	10 days 28 days	14 days a fewdays
22	26	F	Right base anteriorly	9 days	10 days

Table No. 5

Table showing cases with friction rubs and the situation, time of onset and duration of the rubs.

Case No.	Pain and Friction Rub Relationship.
5	Pain and rub appeared together Recurrence of both pain and rub noteworthy
10	Pain entirely gone and patient symptom free when rub appeared.
12	Pain entirely gone when rub detected
21	Pain and rub appeared together Recurrence of rub at different site when had no pain and was feeling well
22	Pain entirely gone when rub detected

Table No. 6

Table showing relationship of pain and friction rubs.

Special Investigations.

X-Ray

Radiological examination of the chest was carried out on all patients who had friction rubs and several others in the series, and no abnormalities were noted.

Duration of Illness.

More than half of the patients were free from pain in a few days, and were comparatively well within a week. Others were unwell for two weeks, and a few especially those with friction rubs took a longer period to recover. All eventually cleared up with no remaining sequelae.

Complications.

No complications were noted. One patient, case No. 22 was four months pregnant when she was affected. The baby has been born and is normal in every respect. She was beyond the time when the foetus is attacked by virus infections.

Enquiries were made from the Medical Officer of Health for the area, and from the Superintendent of the local Infectious Diseases Hospital, and no associated outbreak of Benign Lymphocytic Meningitis was reported.

Polio-myelitis or abortive cases were not encountered in the district at the time of the outbreak.

Differential Diagnosis.

Pneumonia was excluded by the absence of abnormal clinical signs in the chest and by the subsequent course of the illness.

Pleurisy (Tuberculous). All cases with friction rubs were X- Rayed, some of them several times, and no abnormalities were noted on the plates.

Abdominal conditions were ruled out by the absence of typical signs and symptoms, and by the fact that the pain in most cases was related to respiration. When the epidemic nature of the condition was recognised, the diagnosis became much more easy.

Recurrences.

Each patient was reviewed in the Autumn of 1951 and the Autumn of 1952, and the recurrences noted. In Autumn 1951, three patients reported slight pain for one day 2 - 3 weeks after the initial attack (Nos. 11, 19, & 20). Two patients (Nos. 1 & 4) had recurrence of pain when they were still being attended, this taking place 2 - 3 weeks after the onset of the illness. Thus five patients had this type of recurrence 2 - 3 weeks after the initial attack. During the year Autumn 1951 to Autumn 1952, four patients had recurrence of pain, as noted.

Case No. 1.

This boy was said to have had pains every week.

He was admitted to hospital for investigation in June 1952, and had an operation for removal of appendix and Meckel's diverticulum. As stated in his case history no abnormality was found, and the pain has not recurred since then.

Case No. 3

This man had two very slight attacks during the year.

Case No. 12.

This man sought advice while having an attack in September 1952. His symptoms and signs were typical but much less severe than in the first attack. Radiological findings were again negative. He gave a history of three slight attacks during the year.

Case No. 19.

This man had a very slight recurrence in August, 1952.

Prognosis.

As previously stated all symptoms cleared up and no sequelae were left, except the possibility of a slight recurrence.

Treatment.

When first seen, more than half of the cases were given sulphatriad, until the possibility of pneumonia was ruled out. On the non-appearance of definite signs,

the drug was usually discontinued after a very short course.

Otherwise treatment was symptomatic.

Discussion.

In this section I wish to take each point dealt with in the last section, record the views of other observers and compare and contrast my own findings with theirs.

Location.

During the past 20 years, outbreaks of Bornholm disease have been recorded in most parts of the world. Some of these outbreaks have been widespread e.g., U.S.A. outbreaks; others have been more localised. Outbreaks in schools and colleges have been reported by Attlee et alii (1924), Carters (1933), Locke & Farnworth (1936) and Craig (1950).
Greene (1924) reported an outbreak in an orphanage. Akel (1944) had cases in a military camp, and Huss (1934 -cited by Scadding 1946) reported an outbreak on a warship.
Hospitals and convalescent homes have also been affected - Williamson (1924), Lloyd (1924), Welborn (1936) and McConnell (1945). Sylvest (1934) described an outbreak on the island of Bornholm and McDaniel (1944) an outbreak on a small Caribbean island.

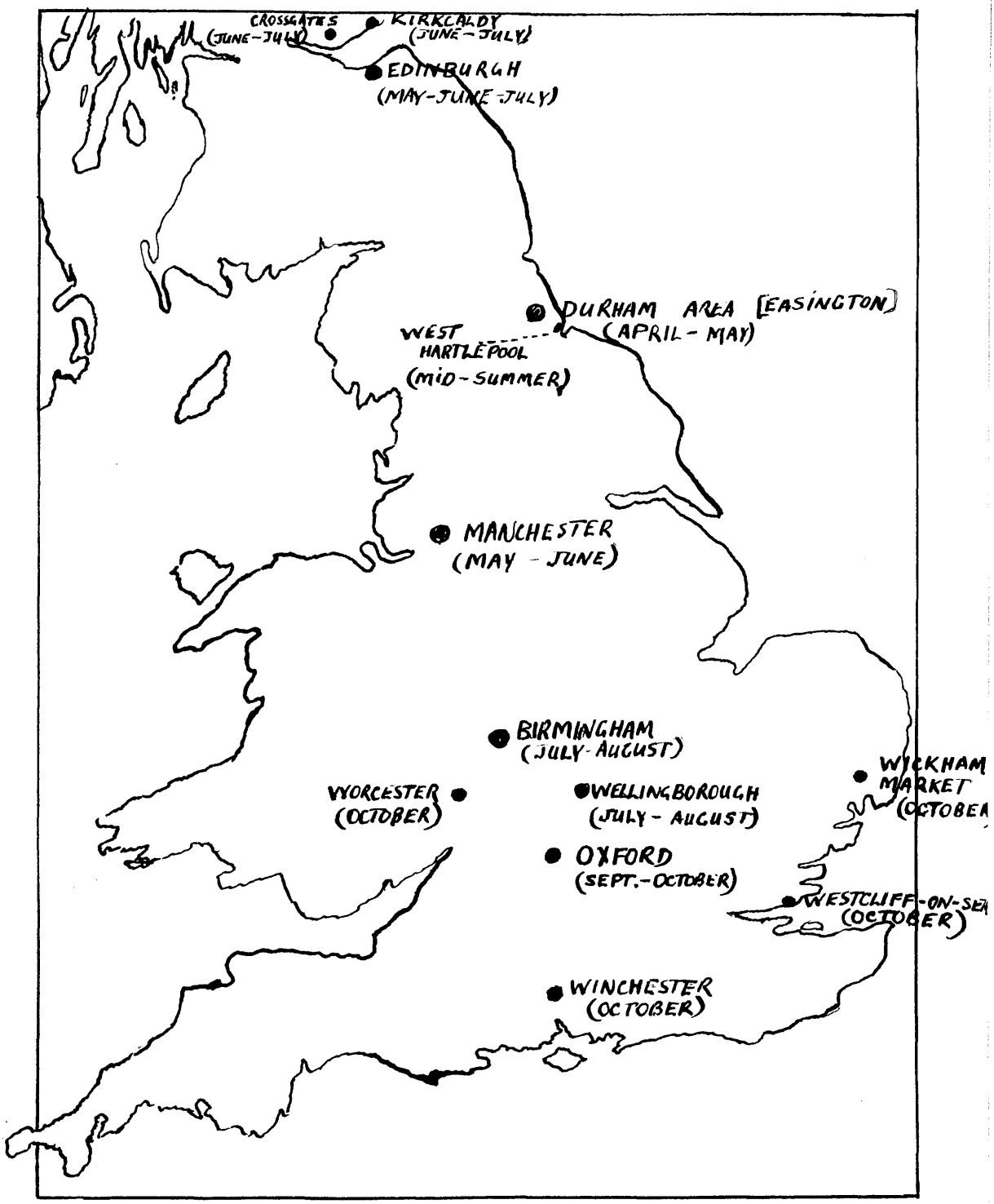
In the Summer and Autumn of 1951, Bornholm disease was prevalent in various parts of Britain. Map No. 2, shows the areas in England and Scotland from which cases were reported in the leading medical journals. Cuneen (1951) also reported cases in Ireland.

Incidence.

The earliest cases in Britain in 1951 were in the Easington Colliery district of Durham (Brown & Prinsley 1951). These cases occurred in the months of April and May. From there the epidemic probably spread northwards to the Forth area. Cases were reported in Edinburgh (Mitchell 1951) and Kirkaldy (Sandilands 1951) at the same time as those seen in Crossgates. It may also have been spread from there southwards to the Manchester district, as cases were recorded in Manchester (Brown et alii 1952) at the same time as the Forth outbreak. The towns then affected are shown in Map No. 2, and were as follows:-

West Hartlepool	(Brown & Prinsley 1951)
Birmingham	(Pinsent 1951)
Wellingborough	(Walton 1951)
Oxford	(Davis & Warin 1951)
Worcester	(Lewis 1951)
Wickham - Market	(Cruikshank 1951)
Westcliff-on-sea	(Quinn 1951)
Winchester	(Swift & Heatley 1951)

AREAS REPORTING BORNHOLM DISEASE IN 1951.



MAP No. 2.

It was the month of October before cases were being reported from the South of England. The Summer and Autumn months would therefore appear to be the time of year when Bornholm disease occurs in Britain, but cases must occur later in the year as well. Tidy (1942 and 1951) reported cases which he had seen when serving with H.M.Forces, and these cases occurred in the late months of 1941 and early months of 1942. During the winter of 1950 Stewart (1951) thought she had seen cases in London. Nevertheless the summer and autumn months are considered by most observers to be the seasons when the disease is most prevalent. Pickles(1933-1939) recorded his own cases during these seasons and quoted Lindberg (1934) writing in Acta Paediatrica, as having had 80 cases from the middle of August to the middle of October. Scadding (1946) in the Middle East, Hamburger and McNeil (1947) in India, and Nichamin (1945) and Finn et alii (1949) in U.S.A. all report their outbreaks in the summer and autumn months. It was also pointed out by Sylvest (1934) that in 1896, Douglas recorded an epidemic of pleurisy (probably Bornholm disease) in New Zealand in the months of February and March. These months correspond to the August and September of the Northern Hemisphere. The cases recorded by Akel (1944) also occurred in the Summer months of the Southern Hemisphere.

Weather.

The type of weather prevailing at the time of the epidemics has been the subject of comment by only a few observers.

Dabney (1888) noted that it was much cooler than it should have been for the season of the year, whereas Harder (1936) had the impression that there was a little more rain than usual, and that the average temperature was a little above normal. Excessive rainfall with periods of continuously high humidity were also noted by Nichamin (1945), and Hamburger & McNeil (1947) described long periods of moist heat.

The weather during the Crossgates outbreak has already been stated, and the heavy rainfall just before the peak period, noted.

Infectivity.

The fact that the disease is contagious has been recognised for many years, but Dabney (1888) considered that it was far less so than most contagious diseases. Payne & Armstrong (1923), however, noted a high incidence in members of one house-hold, as did Sylvest (1934) and Hopkins (1950). Davis & Warin (1951) also stated that it was "the rule rather than the exception for all the children in a house-hold to be involved". The series recorded in Crossgates did not show this high incidence in the families affected. This would indicate that

the disease was probably not so infectious as it appeared to be in other outbreaks. A similar experience was recorded by Nichamin (1945). He reported that in his series there was frequently only one person in the house-hold affected.

Contact at work, which was a probable cause of spread in Crossgates, has been considered to be an important factor in some of the wide-spread outbreaks. The Easington pit disaster (May 1951) was blamed for the spread of the disease in Durham, as many of the relatives of the miners came into the district when the outbreak was at its height, and returned to their homes carrying the infection with them (Brown & Prinsley 1951).

The exact mode of transmitting the infection is still unknown. Ronald (1942) considered that there was no evidence of transmission by food, water or animal vectors, and Jamieson & Prinsley (1947) were of the opinion that the disease was probably spread by droplet infection. Now that the Coxsackie virus has been proved to be the causal agent, these views may require to be altered. Melnick et alii (1950) have isolated the Coxsackie virus from sewage and flies, and Sylvester (1950) has suggested an alimentary spread.

Incubation Period.

Most observers are of the opinion that the incubation period is short. It has been given as 1 - 3

weeks by Nichamin (1945), and 2 - 19 days by Sylvester (1950), but other writers give it as under 1 week e.g. Sylvest (1934) 2 - 4 days, Pickles (1939) - 4 days, Ronald (1942) 2 - 4 days, Hopkins (1950) 4 - 8 days, Davis & Warin (1951) 4 - 5 days. During the epidemic in Britain in 1951, the figures given for the Crossgates series, (viz 3 - 6 days) appeared to be fairly typical.

Duration of Infectivity.

The duration of infectivity has not been the subject of much comment in the literature. Pickles (1933 - 1939) considered it to be brief. It is probably up to the end of the acute phase of the illness.

Age

The greatest incidence of the disease appears to be in those under 30 years of age. Nichamin (1945) had 68% under 30 years, in his series, and Finn et alii (1949) reported 80% under 30 years of age. Ronald (1942) thought there were more cases in the 15 - 30 years group, but Sylvest (1934) stated that the disease was one and a half times more frequent in children under 15 years of age, than in adults over 15 years of age. In the series in Crossgates, the figure of 70% under 30 years of age compares with those given above, but the other figure of 20% under 15 years of age is not like that experienced in other parts of Britain in 1951.

Davis & Warin (1951) had 66 children in their series of 79 - 83%. The difference between the two figures may have been due to the fact, that as previously stated, the disease in Crossgates appeared to be less infectious in families than it was in the Oxford outbreak.

Sex.

The distribution between the sexes has been stated to be equal by Sylvest (1934) and Harder (1936), but Ronald (1942) was of the opinion that males were more often affected, and in the series reported by Nichamin (1945), there was a slight pre-ponderance of females. As mentioned beforehand, the figures for Crossgates were 17 males and 7 females. These apparently support Ronald's view, but the miners who were probably infected at work may have caused some of the difference between the sexes.

Prodromata.

Prodromata are usually considered to be rare. A feeling of chilliness and headache before the onset of the illness were described by a few of Payne & Armstrong's (1923) patients, and Locke and Farnworth (1936) and later McDaniel (1944) reported the complaint of malaise for some hours. Welborn (1936) also had patients with vague symptoms before the illness e.g. a feeling of tiredness for 2 - 7 days before the onset, or a vague generalised

abdominal cramp not associated with nausea, vomiting or diarrhoea. Only two of the Crossgates patients had a feeling of being "run down" prior to the illness.

Onset.

As a rule the onset is abrupt with pain as the outstanding feature. Even in one of the early accounts of the disease given by Dabney (1888), pain was described as the first symptom, preceding the elevation of temperature by half an hour. This abrupt onset with pain was demonstrated in the Crossgates cases.

Signs and Symptoms.

Pain.

The site of pain has been discussed by many writers. Typical reports are as follows:- Locke and Farnworth (1936)- The localisation of the pain at the diaphragm level is typical of the condition.

Akel (1944) - 73% of the cases had pain in the upper abdomen, 44% being in the right quadrant. 26% had pain in the thorax.

Davis & Warin (1951) - 42 out of a series of 79 patients had pain in the abdomen, 11 had pain in the thorax, and 22 had pain in both abdomen and thorax.

Lazarus et alii (1952) - 64% of the series had abdominal pain and 23% thoracic. As can be seen from the above figures and those of the Crossgates series, abdominal

pain occurs much more frequently than thoracic. In children, abdominal pain is almost always present. This was the case in Rector's series (1935). Hopkins (1950) also reported this, and Davis & Warin (1951) stated that abdominal pain which occurred more frequently among children than adults, began in the umbilical area and moved upwards. This may be due to the fact that children use their abdominal muscles in breathing, more than adults do. Sylvest (1934) referred to the frequency with which the right side was involved in his series. He quoted Greene (1924) as finding the left side more often affected, and Josephson (1931) as stating that the two sides were involved with almost the same frequency. In the Crossgates series, the abdominal pain appeared to be more on the right side, and the thoracic pain on the left. The fact that the pain was related to deep breathing was stressed by Hopkins (1950). This was also seen in the Crossgates cases. Hopkins considered this to be an important point in the differential diagnosis. Other factors which aggravate the pain are active movement of the body, (Payne & Armstrong 1923), and sneezing, laughing, or anything which causes movement of the diaphragm (Locke & Farnworth 1936). As previously stated movement of the trunk and walking increased the pain in some of the Crossgates patients. The adjectives

used to describe the pain in this series, as given on a previous page give an indication of the severity of the pain at the onset of the illness. Locke & Farnworth (1936) described the pain at this stage as overwhelming. Another feature of the pain is its spasmodic or intermittent character. As Davis & Warin (1951) have pointed out, young children usually play quite happily between the spasms. This feature of the pain allowed some of the male patients in Crossgates to continue at work for a few days at the commencement of the illness.

Relapses.

Not only is the pain spasmodic and intermittent, but the condition on a whole is spasmodic or relapsing. There is usually a period when the patient is apparently free from symptoms, which is followed by an attack, similar to, or of less severity than the first attack. Exertion or exposure was thought to be the factor initiating the relapse by Payne & Armstrong (1923). The number of relapses have been counted by several observers and Locke & Farnworth (1936) divided their cases into mild or severe according to whether they had relapses or not. In the series reported by Scadding (1946), six of the patients had relapses, and in two of these six, pain contra-lateral to the initial pain, appeared 2 - 3 days after the original one.

Dabney (1888) also referred to the pain changing from side to side in a few of his patients. The relapsing nature of the illness and the appearance of the pain on the opposite side during a relapse, was demonstrated in the Crossgates series, as mentioned before.

Referred Pain.

The fact that pain can be referred from the diaphragm has been dealt with in a previous section. Various authors have given supporting evidence as stated below.

Crone & Chapman (1933) - Three of their patients had right shoulder pain as well as abdominal pain, and two had slight stiffness of the neck.

McDaniel (1944) - In almost all the patients there was shoulder pain at one time or other, and this pain was not increased by movement, except on breathing.

Scadding (1946) - One patient had pain in the left shoulder.

Jamieson & Prinsley (1947) - Bilateral shoulder-tip pain was present in 3 of the series and unilateral in 1.

Lazarus et alii (1952) - 20% of the patients had stiff necks.

It was the opinion of Locke & Farnworth (1936) that referred pain could affect almost anywhere on the trunk e.g. lower or lateral abdomen, lower back, upper border

of trapezius, side of neck, front of chest and tip of shoulder. Reference of pain to the abdomen, the neck and the shoulder tip was demonstrated in the Crossgates series.

Gastro-Intestinal Symptoms.

Gastro-intestinal symptoms are not outstanding symptoms in most descriptions of the disease. Pickles (1933, 1939) stressed the absence of these symptoms - especially vomiting, and Sylvest (1934) stated that seldom had he seen it. In the Oxford outbreak, Davis & Warin (1951) found that vomiting was not common in their series, and only occurred during the most acute attacks of pain. On the other hand, Payne & Armstrong (1923) reported that one-third of their patients had vomiting and that constipation occurred more often than diarrhoea.

Vomiting at the onset was reported by Nichamin (1945) in 40% of his cases, and by Mitchell (1951) in 28%. Lazarus et alii (1952) also reported nausea and, or vomiting in 59% of their series and of the 30 patients reported on by Brown et alii (1952), 12 had vomiting, 8 had diarrhoea and 4 were constipated.

Vomiting at the onset was noted on only two occasions in the Crossgates series, and only one patient complained of constipation. From the figures given above it will be seen that vomiting does occur in some

outbreaks, and not so often or not at all in others. It appears to be more common in the American outbreaks (e.g. Nichamin and Lazarus et alii). The onset of the illness, when the patient is having acute spasms of pain, appears to be the most common time for its occurrence. Diarrhoea and constipation are met with, but are comparatively rare.

Headache.

Unlike the gastro-intestinal symptoms, headache as a symptom is found in almost all recorded outbreaks. It was regarded as one of the three cardinal symptoms by Locke & Farnworth (1936), the others being pain and fever. McDaniel (1944) had 50% of his series with frontal headache, Nichamin (1945) 57%, Akel (1945) 48%, and all of the cases reported by McConnell (1945) had severe headache. The occurrence of headache at the commencement of the illness was referred to by Jamieson & Prinsley (1947) but Davis & Warin (1951) recorded headache 3 - 4 days after the onset, and in their cases where the headache was severe, photophobia and aching of the eyes were prominent symptoms. In the Crossgates patients, headache occurred at the commencement of the illness in 9 of the 24 patients. Headache would thus appear to be a fairly common symptom, and the frontal region appears to be the most common site.

Fauces.

The complaint of sore throat and the appearance of the throat seems to vary in different outbreaks. Attlee et alii (1924) described a red granular pharyngitis, whereas Pickles (1933, 1939) and Sylvest (1934) stated that redness of the throat was rarely ever seen. Howard et alii (1943) reported congestion of the pharynx in all the infants, and three-quarters of the older patients in their series. They cited Fulghum (1942) as reporting that all his patients had pharyngitis. Mitchell (1951) stated that 6 of the 18 children in his series had slight faucial injection and Lazarus et alii (1952) and Brown et alii (1952) also reported sore throats in 39% and 53% of their cases. In the Crossgates series as previously stated, sore throats and faucial injection were found in only 4 of the patients. Throat inflammation is therefore not an outstanding feature in Bornholm disease, but it does occur, and this is understandable when it is remembered that epidemics of sore throats - Herpangina - may be caused by the Coxsackie Virus.
(Von-Magnus - International Congress of Medicine 1952).

Temperature.

The range of temperature recorded in the Crossgates series was much the same as that reported by other observers.

Davis & Warin (1951) gave their figures as 102°F. - 103°F. (38.8°C. - 39.4°C.), and Mitchell (1951) recorded that the usual temperature was 99°F. - 100°F. (37.2° - 37.8°C.), and that in no case did the temperature exceed 101°F. (38.3°C.).

As previously stated Dabney (1888) considered that the temperature rose half-an-hour after the onset of the pain. Locke & Farnworth (1936), on the other hand, thought that the fever came along with the pain, and that in the absence of complications it dropped to normal in 24 - 48 hours, but with each relapse there was another rise. Another observer of this "pain fever" relationship was Welborn (1936). He considered that fever usually followed pain, there being a lag of a few hours, while the reverse was true at the end of a paroxysm.

The rise and fall of the temperature with the pain of a relapse was seen in some of the Crossgates patients. This appears to be a definite feature of the condition.

Sweating.

The occurrence of sweating when the temperature is coming down was described as early as 1888 by Dabney. It was also reported by Payne & Armstrong (1923), and Rector (1935) also referred to it. In the report by Swift & Heatley (1951) it was stated that "the lowering of the temperature was accompanied by a drenching sweat,

during which the patient felt exhausted and anxious". Five of the Crossgates patients also had this complaint.

Lymphadenopathy.

Enlargement of the lymph glands was not noted in any of the Crossgates patients, but reference is made to it here since it has been recorded elsewhere. Finn et alii (1949) reported lymphadenopathy in 45 of their series of 114 patients, and stated that they had not seen it previously described. One of the cases reported by Hopkins (1950) had enlargement of the lymph glands and had actually been considered to be a case of Glandular Fever, until a Paul-Bunnel test was declared negative.

Abdominal Examination.

(a) Tenderness.

Upper abdominal tenderness is met with fairly frequently. Sylvest (1934) considered that the tenderness was in the muscles of the abdominal wall, but could not localise the tenderness there to any definite muscle alone. In his patients it occurred more on the right side than on the left. He also noted swelling of the muscles e.g. rectus abdominis. Another view was expressed by Locke & Farnworth (1936) They thought that in the areas where there was pain, the

skin was hyperaesthetic and that there was only slight muscle tenderness. When such areas were examined, there might be muscle spasm, but it seemed to them that the tenderness was largely confined to the skin. Akel (1944) compared the sites of tenderness with those of the pain, and found them much the same. Forty percent of his patients were tender in the right upper quadrant, 8% in the left and 15% in the epigastrium.

Other observers who reported upper abdominal tenderness, more often on the right side, were Scadding (1946) and Finn et alii (1949). This was also seen in the Crossgates patients, but no muscle swelling or tenderness confined to certain muscles was observed.

(b) Guarding.

Guarding of the abdominal muscles is seen occasionally. It was noted in one of the Crossgates patients. Jamieson & Prinsley (1947) reported 4 of their series of 35, who had upper abdominal guarding and rigidity. The combination of pain, tenderness and guarding or rigidity has led to the diagnosis of an abdominal emergency on several occasions. Payne & Armstrong (1923) had one such case and Crone & Chapman (1935) reported two cases who had exploratory laparotomies performed with the primary diagnosis of appendicitis, and another one with a previous diagnosis

of perforated duodenal ulcer. Five cases which simulated abdominal emergencies were seen by Hamburger & McNeil (1947), and a very unusual case with symptoms and signs simulating ruptured ectopic pregnancy was recorded by Prisman & Shrand (1950).

(c) Referred Pain from Abdominal Palpation or Pressure.

Deep palpation in the right hypochondrium produced pain in the right shoulder in one case in the Crossgates series.
This was also reported by Scadding (1946). The explanation must be that the abdominal contents are pushed against the tender central portion of the diaphragm, giving rise to reflex pain through the phrenic nerve. Craig (1950) found that pressure on the upper abdomen produced pain in the infra-clavicular region - another example of referred pain.

(d) Abdominal Reflexes.

Abnormality of the upper abdominal reflexes has been reported, but this will be dealt with in a later section on the neurological complications of Bornholm disease.

Chest Examination.

Thoracic tenderness is not so common as abdominal

tenderness. The figures given by Akel (1944) viz. 4% on right side of thorax, and 7% on left are typical of the frequency of this finding. In the Crossgates series it was found in only one case. Finn et alii (1949) noted signs of respiratory tract involvement in a few of their patients, but this was not seen in Crossgates and is seldom referred to in the literature. Pleural friction occurs much more frequently. Among the earliest accounts in this country were those by Williamson (1924), Lloyd (1924) and Attlee et alii (1924). Williamson described cases of what he termed "epidemic pleurisy". They occurred in a children's Hospital. In his series the friction rubs were detected early in the illness - usually on the second day, and were no longer audible by the end of the week. Lloyd's cases were also in a Children's Hospital, but were among the nursing staff, and the friction rubs were heard later in the illness. The other outbreak in 1924 which was described by Attlee et alii occurred among boys in their teens. Seventeen of their series of 48 had friction rubs which were loud and could be felt or heard by the patient long after the acute pain had gone. Other authors who reported pleural signs were Sylvest (1934) who had two cases, and Huss (1934) - cited by Scadding (1946) who had 20 cases, but one of the most interesting reports was given by Locke & Farnworth (1936).

They stated that in only one of their series of 121 did they find pleural signs during the early days. A faint pleural friction rub was present over a small area for a few hours. Later in the illness seventeen patients developed friction sounds which they considered indicated the presence of "fibrinous pleuritis". These sounds were present for a period of one to several weeks, and were of a coarse grating character and of extraordinary intensity. They could be heard over an extensive area and could be palpated as well as heard. Very often the patients felt the rubs themselves, and none of them had any pain when the rubs were present. Another interesting account of friction rubs was given by Scadding (1946). He had eleven patients who had rubs 3 - 9 days after the onset of symptoms. Five of these patients had rubs on both sides of the chest - unilateral at first and appearing on the second side 1 - 6 days after the appearance on the first side. These friction rubs were always at the extreme base and in 5 cases were palpable. They often appeared after the most severe pain abated, and sometimes persisted after the actual pain had disappeared. A few patients who still showed palpable friction felt quite well, with only a slight dragging sensation on deep inspiration. Three of the 5 patients with bilateral friction had no pain on the side on which the second friction sound appeared.

Jamieson & Prinsley (1947) and Finn et alii (1949) also reported friction rubs and Hopkins (1950) remarked on the "curious lack of association between the severity "of the pain and the loudness of the rub". Pleural friction rubs during the first few days of illness were found by Sylvester (1950) in two cases. In one of them an X-Ray plate of the chest taken on the second day of illness showed a slight but definite decrease of translucency at the right base. This was more marked on the 4th day of illness and was associated with slight elevation of the right half of the diaphragm. Sylvester considered these X-Ray findings to be due to pleural oedema.

In 1951 Tidy commented at length on the friction rubs found in Bornholm disease. He referred to a series he had seen when in H.M. Forces, and to a previous account given in 1942. I now quote from his letter to the British Medical Journal.

"Pain is caused by movement and is not closely related "to the rub when this is present. The rub may persist "after the pain has subsided, or develop on the opposite "side without any pain. It may be extremely loud and "coarse, and of a character which I have never heard in "pleurisy, although in other instances it may be "indistinguishable". He thought that the rub was not a pleural rub, but of muscular origin, possibly from the sheath or tendon. All of the cases with friction rubs in the Crossgates series were adults (Table No. 5).

Three were over 30 years of age and two in the 15 - 30 age group. In the cases reported by Davis & Warin (1951) there were 66 children (out of 79 cases), and no friction rubs were noted. Mitchell (1951) who saw cases at the Royal Hospital for Sick Children, Edinburgh, also reported no friction rubs.

From these facts one is tempted to conclude that friction rubs only occur in adults. If Tidy's hypothesis is correct and the friction rubs arise in the muscles, then one should only look for them in adults, since, as already mentioned children do not use their thoracic muscles when breathing to the same degree as adults. Against this theory is the fact that Williamson (1924), as stated above, reported definite pleural friction rubs in the first few days of illness in his series in children. This leads one to suppose that there might be two kinds of friction rub met with in Bornholm disease - (1) a rub which is due to muscle friction and does not occur or is seldom found in children and (2) the true pleural friction rub which can occur in both children and adults. That this supposition may be correct is borne out by certain other characteristics of the rubs which will now be analysed. In the Crossgates series the rubs were detected on the 8th - 10th day of illness in four of the five patients (Table No. 5). It will thus be seen that the rubs appeared comparatively late in the illness, and they had a prolonged course. On going over the findings of the writers quoted above, one finds that

most of the rubs reported were late also, except for the following, (1) Williamson (1924). This observer, as previously stated found friction rubs on the second day. (2) Locke & Farnworth (1936). They had one case with pleural signs in the early days. (3) Sylvester (1950). He had friction sounds and radiological evidence of pleural involvement on the second day of illness. The ones which occurred early would appear to have been pleural in origin, as Sylvester had X-Ray proof, the ones which occurred late were probably muscular in origin. Locke & Farnworth (1936) as already quoted, considered that the late rubs were due to fibrinous pleuritis, but if the above supposition is correct, they would be due to muscular friction. Table No. 5 shows that the friction rubs in the Crossgates series were present for several days up to 14 days. In the series reported by Williamson (1924) and Sylvester (1950) - considered to be cases of true pleurisy, the rubs were detected for two days only. Table No. 5 also gives the site of the rubs. As can be seen, all of them occurred at the base and most of them anteriorly. It has been pointed out by Bray (1926) that the ribs separate appreciably at the base, but not at the summit of the chest. Anteriorly at the base of the chest and in the axilla, their separation is greater than posteriorly. There is therefore more movement of the intercostal muscles at the base anteriorly, and this is

the site where friction sounds are most frequently heard. The fact that the fibres of the internal and external intercostal muscles run at right angles to one another may be a factor in the causation of the rubs. As has been already stated, the late friction rubs reported by Locke & Farnworth (1936) were heard over an extensive area. The true pleural friction rubs, on the other hand, although they also occur most often at the bases, appear to be much more localised. Williamson (1924) has stated that "the area over which the friction "rub was heard was generally confined to a very "circumscribed area at the base". Examination of Table No. 6, will show the relationship of the rubs and the pain. The frequent occurrence of the rub when there was no pain, and the recurrence of the rub at a different site when there was no pain, were features of the cases. This characteristic has been noted by other observers as already indicated. It would appear to suggest muscle origin, as in true pleural friction, the pain and friction rubs appear almost together e.g. Sylvester's cases. It would also appear to suggest that resolution was beginning to take place in the inflammatory process. The stage of oedema and swelling of the muscle (which is the painful stage) was passing and absorption was taking place. As a result friction between the fibres of the muscles or between the muscles themselves was taking place and giving rise to friction sounds.

The coarse palpable character of the friction rub was noted in one of the Crossgates patients. This has also been referred to by other authors, and would point to muscle origin, rather than pleural origin, where the friction sound is much fainter. The characteristics of the two friction rubs would thus appear to be as follows:-

(1) Muscle friction rubs.

These rubs occur in adults - probably due to the different type of breathing. They occur late in the illness and very often last for some time. Their commonest site is at the base anteriorly - due to the movement of muscles, as has been shown, but they are sometimes heard over an extensive area. They usually occur when the acute phase of pain has passed or is passing, and very often when there is no pain. They are coarse, loud, grating, and often palpable, and the patient can feel or hear them themselves. On account of the late appearance of the rubs - often when the patient is quite well again, they are probably not so frequently detected as they might be.

(2) Pleural friction rubs.

These rubs occur in both children and adults. They appear early in the illness, usually on the second day, and last only a few days. Their commonest site is at the base, in a localised circumscribed area.

They occur along with the pain and are not loud and coarse, but are faint and more difficult to detect. It will be apparent from these descriptions that the muscle friction rubs are the ones which are more frequently encountered, and they provide corroborative evidence for Tidy's view on the aetiology of the disease. It will also be apparent that true pleural ones occur, but not so often.

As will be shown later, meningitis is now considered to be a complication of the disease. The Coxsackie virus must therefore attack serous surfaces as well as other structures. Whether the pleura is primarily infected or whether it is infected from the diaphragm is a matter on which no definite statement can be made at present. The occurrence of the rub on the second day of illness suggests the former whereas the close attachment of the diaphragmatic pleura to the diaphragm - already referred to, suggests the latter.

Special Investigations.

(a) White blood cell count.

Both leucopenia and leucocytosis have been described.

e.g. leucopenia - Helman & Jaffe (1950)

leucocytosis - Mitchell (1951)

(b) Erythrocyte Sedimentation Rate.

Elevation of the E.S.R. has been reported by the following:-

Nichamin (1945) Scadding (1946)

Helman & Jaffe (1950) and Mitchell (1951)

The only observer who found these two investigations of much value was Mitchell (1951). He stated that he made use of them in differential diagnosis, since the total leucocyte count and E.S.R. were lower than one would expect in such conditions as acute tonsillitis and pneumonia, and therefore these conditions could be ruled out.

(c) X-Ray of Chest.

Radiological examination of the chest has been carried out in several of the outbreaks, but only Scadding (1946) and Sylvester (1950) have reported any abnormalities. Scadding (1946) had fluoroscopy done on 16 of his patients and two, both on the 7th day of illness, showed very slightly restricted movement of the diaphragm on the affected side. Reference has been made on a previous page to the changes described by Sylvester (1950). No changes were found in the Crossgates series.

Duration of Illness.

The duration of the illness is generally considered to be short, except in the most severe cases. When Dabney (1888) described his cases, he mentioned some who had only one paroxysm of pain lasting 12 hours, others who were ill for several days, and two who took three weeks.

He also reported several who had extreme prostration for 2 - 3 weeks after all fever had subsided and who suffered pain in the affected side whenever any unusual exertion was attempted. Attlee et alii (1924) stated that many of the boys in their series took a long time to get well, but this is not usual. Typical figures are as follows:-

Locke & Farnworth (1936) Mild cases 1 - 3 days
severe cases 18 - 19 days

Welborn (1936) 5 days

McConnell (1945) 5 - 10 days

Jamieson & Prinsley (1947) 2 - 7 days

Lazarus et alii (1952) a few days - a few weeks.

The figures for the Crossgates patients were much the same.

Complications.

(a) Pericarditis.

Pericarditis was stated by Sylvest (1934) to have been reported by physicians in the Skien outbreak in 1896. One case was reported by Payne & Armstrong (1923), three by Locke & Farnworth (1936), and one by Finn et alii (1949). It is therefore a very uncommon complication.

(b) Orchitis.

Orchitis associated with Bornholm disease was seen by Sylvest (1934), in one patient only, but Huss (1934) - cited by Scadding (1946), reported 50 cases in his series.

Jamieson & Prinsley (1947) described 12 of their 30 patients as having had this complication. It was usually unilateral, affecting each side equally, and came on 8 - 39 days from the onset of the illness. Two cases were also reported by Finn et alii (1949).

(c) Pneumonia.

One case of "mild broncho pneumonia" was recorded by Locke & Farnworth (1936), and Sylvest (1934) quoted other observers as having seen cases. It must be very rare, however, and when it does occur, it is probably a coincidence.

(d) Otitis.

Otitis was described as a complication by Payne & Armstrong (1923), and again by Nichamin (1945) but it, also, is very uncommon.

(e) Pleurisy.

This has been fully described in a previous page.

(f) Skin Rashes.

Crone & Chapman (1933) recorded a scattered discrete maculo - papula punctate lesion of the skin - said to be like mosquito bites, and Williamson (1924) a sudaminal rash appearing when the temperature fell. No other references to skin lesions have been found in the literature.

(g) Urinary Complications.

Urinary frequency was described by Walton (1951) in some of his cases. Catheter specimens showed no bacteria, pus cells or albumin.

(h) Neurological Complications.

Among the first references to the neurological complications of Bornholm disease was the account given by Sylvest (1934), of abnormality of the upper abdominal reflexes. He also cited a Danish physician as thinking he had observed encephalitis in association with the disease. Huss (1934), quoted by Scadding (1946) reported one case of meningitis or meningo-encephalitis, and Lindberg (1934) also quoted by Scadding, 5 cases. Other reports may be summarised thus:-

Howard et alii (1943). They reported 5 adults and 1 child with meningo-encephalitis, and several of the children in their series had convulsions.

McConnell (1945). Of the 16 patients in this series, all had severe headaches, 13 had hyperaesthesia referred to the thorax and legs; 12 had photophobia and or nuchal rigidity and one had a positive Kernig's sign: three had symptoms resembling those of meningo-encephalitis. McConnell also quoted previous authors who had drawn attention to Benign Lymphocytic Meningitis occurring concomitantly with Bornholm disease in Cincinnati in 1935.

Nichamin (1945) :- Paraesthesia in some of the female patients was reported. 40% of the whole series complained of giddiness.

Hamburger & McNeil (1947):- They noted the co-existence of an outbreak of Lymphocytic Meningitis at the same time as their outbreak of Bornholm disease.

Finn et alii (1949):- Abnormal cerebro-spinal fluid was recorded in 4 of the 18 patients who had lumbar-puncture done.

Davis & Warin (1951):- Four cases in whom meningitis symptoms were predominant were reported. They all had some abdominal or thoracic pain as well, and their contacts had typical attacks of Bornholm disease.

Brown & Prinsley (1951):- These observers stated that in addition to a widespread epidemic of Bornholm disease in Durham in 1951, there were sporadic cases of Benign Lymphocytic Meningitis.

Gsell (1950 - cited by Thelin & Wirth 1951):-
An account of 7 cases of serous meningitis in association with Bornholm disease was published, and the term "meningitis myalgica" coined.

Thelin & Wirth (1951):- These authors considered that in the meningeal form the muscular pains are variable and, or even absent, just as the paralysis may be absent in the meningeal form of poliomyelitis.

Only one of the Crossgates patients showed transitory photophobia and meningeal signs, but the reports quoted

leave no doubt in one's mind that meningitis or meningo-encephalitis is a definite complication of Bornholm disease.

Dalldorf (1950) reported lesions in the brain as well as in the muscles of suckling mice injected with the virus, so it may be that the primary lesion is an encephalitis and the meninges are infected secondarily. Further study of this problem is necessary especially in the groups of cases which hitherto have been classified as Benign Lymphocytic Meningitis, Aseptic Meningitis, or even Non-paralytic Poliomyelitis.

Differential Diagnosis.

(a) Chest diseases.

Tuberculosis, Pleurisy and Pneumonia must be excluded. McDaniel (1944), reported that one of his cases was sent to hospital with a previous diagnosis of coronary thrombosis, and Mitchell (1951) also referred to one of his series being labelled a doubtful angina. Heart disease must therefore also be considered.

(b) Abdominal diseases.

The number of times laparotomy has been performed has been mentioned in a previous section. Abdominal emergencies must always be borne in mind. Cholecystitis is another condition which may cause some difficulty.

This was seen in one of the Crossgates patients.

(c) General Diseases.

Influenza and Glandular Fever have been included in the essential differential diagnosis by Finn et alii (1949).

(d) C.N.S. Diseases.

Non-paralytic poliomyelitis and lymphocytic meningitis due to other causes, should be excluded.

Recurrences.

Recurrences of pain were described in the account of the Oxford outbreak by Davis & Warin (1951). These recurrences were experienced a short time after the initial attack, and in the case of children they seemed to occur when they returned to school too early. Reference has already been made to the relapsing nature of the illness. The recurrences described by these authors would appear to be an extension of the relapses. Five of the Crossgates patients had recurrences 2 - 3 weeks after the initial attack. All were of much less severity. In 1952 Nichamin published an account of 17 patients who had had recurrences at intervals of 2 - 30 months. Some had had only one recurrence, others two, three or more. He stated that there was little relationship between the recurrences and the season

of the year, and that they occurred in the younger patients as well as the older ones. He considered that the repeated attacks of this infection in the same person resembles other disease conditions with a virus aetiology e.g. Herpes Simplex. "The infected person", he wrote, "appears to have an inherited "susceptibility to repeated attacks of the disease, "which are precipitated by various excitant factors". Among the Crossgates patients there were 4 who had attacks during the year Autumn 1951 - Autumn 1952. The first one was a boy who was having recurrent subcostal pain. He was seen by a surgeon who took him into hospital for investigation, and operated, to find no abnormality. The interesting feature about this case is that although no abnormality was found, the pain ceased after the operation. From this finding one is tempted to conclude that there may be a psychological basis to some of the recurrences. Another of these four patients was one who was so incapacitated that he had to stay off work and seek medical aid. When seen he was in the throes of a typical attack of Bornholm disease.

Immunity.

One attack of Bornholm disease does not therefore confer immunity, and as Nichamin (1952) has suggested it may confer a susceptibility to the disease.

Prognosis.

The prognosis is considered by all observers to be good. No fatalities have been reported.

Treatment.

Symptomatic treatment is employed in most outbreaks, but Swift & Heatley (1951) described treatment by chloramphenical, with a "prompt and dramatic response", and Nichamin (1952) used aureomycin, again with very good results. The use of these antibiotics in the severe cases would thus appear to be indicated.

Conclusions.

Bornholm disease is an infectious disease occurring most frequently in the Summer and Autumn months.

It is spread by family contact, house-to-house contact, and contact at work, the actual mode of transmission being unknown.

The degree of infectivity varies in different outbreaks.

The incubation period is short, probably 3 - 6 days.

It is found most often in children and adults under 30.

Prodromata are rare.

The onset in abrupt with pain in the upper abdomen or thorax as the first symptom.

In children the pain is almost always in the abdomen, due probably to the abdominal type of breathing met with at this age.

When the pain is in the abdomen, there is a preponderance of pain in the right side.

Pain is usually aggravated by deep breathing, and frequently by movement of the trunk.

The pain is very severe in most cases and is of a spasmodic character.

Relapses of pain and other manifestations of the

illness are frequent.
Referred pain from the diaphragm is often found.
Gastro-intestinal symptoms are not common, but vomiting at the commencement can occur.
Severe frontal headache is frequently encountered.
Sore throats and injected fauces are occasionally seen.
The temperature range is not high.
A rise and fall of the temperature with the pain of a relapse is a feature of the illness.
Sweating is sometimes seen.
Abdominal tenderness, most often in the right upper region, is another feature.
This tenderness is occasionally accompanied by guarding of the muscles.
The combination of pain, tenderness and guarding may lead to the diagnosis of an abdominal emergency.
Friction rubs may be heard during the course of the illness, and are probably of two kinds, viz.,
(a) Muscle friction rubs.
These rubs have their origin in muscles and occur in adults, probably since adults use their thoracic muscles when breathing. They are found at the base, usually anteriorly, and sometimes over a wide area. It is often late in the illness when they appear, but they remain for some time. Frequently they are unaccompanied by pain, or appear when the pain has gone or is going,

and are of a loud coarse grating nature. They are often palpable and the patients can feel or hear them themselves. Their presence indicates infection of the muscles above the costal margin.

(b) Pleural friction rubs.

These rubs occur in children as well as adults and are found in a circumscribed area at the base, early in the illness. They usually last only a few days and accompany the pain. Unlike the muscle friction rubs they are faint, and difficult to detect. Their presence indicates infection of the pleura by the Coxsackie virus.

As a general rule, radiological examination of the chest shows no abnormality.

The illness is usually of short duration, but some patients are ill for 2 - 3 weeks and others are debilitated for some time afterwards.

Recurrences of the disease occur, but in a milder form than the initial attack.

In assessing the severity of the pain in a recurrence one must bear in mind the possibility of a psychological element.

Summary.

Bornholm disease has been shown to be a much more common disease than is generally known.
A historical outline of the disease has been given.
Evidence has been stated for the assumption that Bornholm disease is due to an infection of the diaphragm, with spread to the muscles above and below the costal margin.
The search for the infecting agent has been described and the evidence that the Coxsackie virus is responsible has been detailed.
Reference has also been made to the similarities between the Coxsackie virus and the Poliomyelitis virus.
The epidemiology and manifestations of an outbreak of Bornholm disease in Crossgates district of Fife, in 1951, have been recorded.
Twenty-four cases have been described.
The apparent spread of the disease in Britain in 1951 has been shown.
Findings in the Crossgates cases have been discussed, compared and contrasted with those of other observers.
The possible complications have been dealt with and

notes on the differential diagnosis, prognosis, and treatment given.

Certain conclusions have been drawn from a study of the cases and literature, and these have been tabulated, special reference being made to the friction rubs which may occur during the course of the illness, and the possible recurrences of the disease.

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