

CARDIAC COMPLICATIONS OF BORNHOLM DISEASE

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THE interesting clinical phenomenon of acute benign pericarditis has been known for over a hundred years (Johnson *et al.* 1961), but it is only in recent years that light has been thrown on its aetiology. It is now known to be a rare complication of a variety of diseases, for example, measles, mumps, poliomyelitis, variola, varicella and Bornholm disease. The Coxsackie Group B of viruses, which are responsible for the latter, can cause both a pericarditis and a myocarditis. There is now sufficient evidence to show that this group can produce a fulminating and often fatal myocarditis in the newborn (Van Grevald and de Jager 1956; Verlinde *et al.* 1956; Woodward *et al.* 1960), but in older children and adults full recovery is the rule (Lewis and Lane 1961). Pericarditis as a complication of Bornholm disease is more common in adults, whilst myocarditis is commoner in children.

This paper describes two patients in the author's practice who had cardiac complications following Bornholm disease. The first occurred in a very large epidemic of this disease in Swansea in 1956 (Williams 1956), and the second in a much smaller outbreak in the same town in 1963.

Case histories

Case 1. The patient, a little girl aged 6½ years, was perfectly well until 23 June 1956 when she developed a sudden severe pain in the right subcostal region, extending into the epigastrium immediately adjacent to the xiphisternum, and a less severe pain in the suprasternal notch. It came on in spasms, causing the child to scream.

She was off her food, listless, feverish and perspired profusely during the periods of pain. Gradually it eased and disappeared towards late afternoon. In the evening the pain returned with great severity, but it was confined to the epigastrium, suprasternal notch and retromanubrial region. After approximately two hours it subsided. There was no headache, nausea, vomiting, diarrhoea, constipation, frequency of micturition nor earache. She had been inoculated against diphtheria three months previously. She had chickenpox when four

years of age and suspected mild thrombocytopenic purpura when she was 5½ years of age. There was no significant family history.

When seen on the first day of her illness, the child was crying because of pain coming on in spasms at intervals of five or ten minutes. Taking a deep breath made it worse or induced it during periods of partial remission. Her temperature was 103°F., and she was perspiring profusely.

Cardiovascular system—the pulse, 100 a minute, was normal. The heart was normal in size, there was no thrill, and sounds I and II were clearly audible in all areas. Examination revealed neither pericardial nor pleuropericardial rub.

The respiration rate was 34 per minute and there was no respiratory grunt. The breath sounds were normal; there was no pleural rub nor muscle creaking. There was some guarding of the upper abdomen and tenderness in the right subcostal region and especially deep to the xiphoid process. The central nervous system was normal.

On the *seventh day* of her illness she began to have periodic attacks of tachycardia and on the *tenth day* the author witnessed an attack which was stopped by carotid pressure. During the attack it was impossible to count the pulse rate. The child was able to stop subsequent episodes by holding her breath. On the 24th day, an electrocardiogram showed a normal tracing with a pulse rate of 70 per minute. A blood count performed on the same day was normal. Coxsackie virus Group B type 3 was identified from the faeces.

The treatment consisted of bed rest, analgesics and, for five weeks, phenobarbitone gr. ¼ three times a day.

The patient continued to have attacks of paroxysmal tachycardia, but they became milder, less frequent, and disappeared altogether after 18 months.

This patient, although the Coxsackie virus Group B type 3 was isolated from the faeces, could have been a carrier for, like the polio virus, it is found in the intestinal tract of a proportion of normal individuals. However, she showed the classical clinical picture of Bornholm disease and had been taken ill during a large outbreak in the town, when it was estimated that over 2,000 cases had occurred. If serological investigations had been carried out, it is likely that a rising titre to the virus recovered from the faeces would have been found. Paroxysmal tachycardia is unusual in children, and it seems to be more than a coincidence that this infrequent occurrence should have happened in association with another uncommon disease in the absence of a common aetiological factor which, in this case, was the Coxsackie virus Group B type 3.

Case 2. The patient, a married man aged 38, was perfectly well until 8 July 1963, when he felt feverish and had a headache. He took two aspirins, went to bed, slept well, and “woke up fine” next morning. The pattern of the *next two days* was the same as that for the first day, except that he had now developed a pain in the right side of his chest. On the *fourth day* he felt well on getting up, but as soon as he exerted himself at work, the pain returned. This time, it was made worse by deep breathing or sighing, and persisted after he went to bed. He perspired a great deal during the night and wisely decided to stay in bed for the following two days. On the *seventh day*, he went to work, but came home in the evening “full of flu”. He had a headache and “ached all over”, but with worse pain in the right side of the chest. On the following day (*eighth day*) breath-

ing became difficult and he was frightened, so he left work early, went to bed, and called the doctor.

When examined, his general condition was good, but he was perspiring a little and had a temperature of 101°F. The respiratory rate was 18 per minute, but deep breathing caused him to grunt, due to a pain in the right subcostal region and the tip of the shoulder on the same side. Breath sounds were vesicular in all areas and there were no adventitious sounds. The pulse was normal at 98 per minute. The heart was not enlarged and there was neither a pericardial nor a pleuropericardial rub. Sounds I and II were clearly audible in all areas. There were no murmurs. There was some guarding of the right upper rectus muscle. The central nervous system was unaffected. Bornholm disease was suspected and a specimen of faeces and blood were sent for virological examination.

On the *ninth day* the pain shifted to the precordial region and was unaffected by breathing. It was now very severe and gave the patient no respite. His pulse rate was 100 per minute. The apex beat was half an inch to the left of the nipple line in the fifth interspace. There was a very definite pericardial friction rub, and a provisional diagnosis of pericarditis following Bornholm disease was made. However, it was important to proceed with further investigations to exclude a more serious cause of pericarditis. He was seen by a consulting physician who admitted him to hospital for investigations. Laboratory tests gave the following results: serum protein, 5.9 mg. per cent; electrophoresis, relative increase in alpha two globulin fraction; blood count, haemoglobin 85 per cent; white cells 5,600 (polymorphs 3,584, lymphocytes 1,736, monocytes 224, eosinophils 56); ESR 40 mm/hour: Serum G.O. transaminase 100 units per cent. X-ray of chest. 17 July showed slight cardiac enlargement and on 2 August it was normal. ECG—17 July—S-T segment significantly raised in leads I, II, avf and especially v7. The ECG on 2 August was normal.

Coxsackie virus Group B type 3 was isolated from the faeces. The first serum specimen showed absence of neutralizing antibody for this virus, but on the 56th day it was present in dilution of 1/64.

The patient was discharged home after a month in hospital and fully recovered, he started work three weeks later.

During the time the patient was taken ill, the author was investigating a small outbreak of Bornholm disease in the town where virological investigations had confirmed Coxsackie virus Group B type 3 as the cause. One of these confirmed cases was a child only 50 yards from the patient's house. The patient's two children had already recovered from a febrile illness which, from the description, was almost certainly Bornholm disease. There was no history of contact between these children and the child living 50 yards away, but there were others in the vicinity with whom they had been playing and who had suffered from a similar illness. Knowledge of the outbreak of Bornholm disease in the town at the time doubtless helped greatly in the diagnosis, for Bornholm disease itself, especially occurring in a sporadic case, can be very difficult to diagnose.

In classical Bornholm disease, prodromal symptoms prior to the onset of pain are uncommon, and, when they do occur, they rarely precede it by more than 72 hours (Williams 1961). This patient's prodromal symptoms preceded the pain by 48 hours. The disease

had affected the right dome of the diaphragm referring the pain to the subcostal region causing some protective muscle spasm. Unlike the pain in the precordial region, which was constant, the diaphragmatic pain varied with the depth of breathing. The changes seen on the electrocardiogram were slight, but confirmed the diagnosis of pericardial involvement.

Discussion

One of the most fascinating problems in Cocksackie virus infection is the contrasting effects in children and adults. Pericarditis is the prominent cardiac complication in adults, whilst in small children, myocarditis is the commoner.

In the newborn, the myocarditis is usually of a fulminating type and many of the children die. This phenomenon also occurs in experimental animals, especially in mice.

An important characteristic of the Cocksackie virus is that, when inoculated into suckling mice, it induces widespread paralysis and death. At post mortem, focal destructive lesions of fat, brain tissue, myocardium and striated muscle are seen. Similar changes in human striated muscle have been demonstrated by Lepine *et al.* (1952) in a Bornholm disease sufferer during the European pandemic outbreak in 1951. Similar changes have been seen in the myocardium of babies who have died of Cocksackie virus myocarditis. The virus has been isolated from the myocardium and also from the pericardial fluid.

Plager and his colleagues (Plager *et al.* 1962) have reported three cases of pericarditis in pregnant women which occurred during an outbreak of Cocksackie Group B type 5 infection in Albany during the summer of 1961. The three patients were delivered of healthy babies.

Case 1 was not as ill as one might have expected in one with acute myocarditis. It is difficult, however, to explain the attacks of paroxysmal tachycardia which started on the seventh day of the illness and persisted for 18 months without considering the possibility of a virus myocarditis. The isolation of the virus from the faeces or throat washings cannot be regarded as conclusive evidence of recent infection. In this case, the circumstantial evidence, which included a classical clinical picture of Bornholm disease, the presence of an epidemic of that disease, and positive virus isolation from the faeces weighed heavily in favour of the diagnosis of Bornholm disease. This case which was only briefly mentioned in a previous paper (Williams 1961) has been included in more detail here, because as far as it can be ascertained from the literature, paroxysmal tachycardia as a complication of Bornholm disease has never been described.

Case 2 showed a clinical picture of Bornholm disease complicated by pericarditis. The virus was isolated from the faeces and serological investigations showed a rising titre to the same virus. A more conclusive proof of the nature of the illness would have been gained from the isolation of the virus from the pericardial fluid, but the opportunity did not arise in this patient.

Summary

Two patients with cardiac complications following Bornholm disease are described. The first developed attacks of paroxysmal tachycardia which lasted 18 months, and the second was complicated by a pericarditis.

Acknowledgements

I wish to express my sincere thanks to Dr William Howell of Swansea Hospital, and to Dr R. T. Jenkins of Morriston Hospital for their valuable help. I am very grateful to Dr A. Evans of Cardiff P.H.L.S. and Dr D. Fleck of Swansea P.H.L.S. for carrying out virological investigations on these cases. My grateful thanks are due to Dr William Evans of the London Hospital for his helpful criticism. Finally, I wish to thank Miss June Johnson for her secretarial assistance.

REFERENCES

- Johnson, R. T., Portnoy, B., Rogers, G. N., and Bueshner, E. L. (1961). *Arch. intern. Med.*, **108**, 823.
- Lepine, P., Derse, C., and Satter, U. (1952). *Bull. Acad. Med. (Paris)*, **136**, 66.
- Lewis, D., and Lane, W. T. (1961). *Lancet*, **2**, 1385.
- Plager, H., Beebe, R., and Miller, J. K. (1962). *Arch. intern. Med.*, **110**, 735.
- Van Grevald, S., and de Jager, H. (1956). *Ann. paediat. (Basel)*, **187**, 100.
- Verlinde, J. D., van Tongeren, H. A. E., and Kret, A. (1956). *Ann. paediat. (Basel)*, **187**, 113.
- Williams, W. O. (1956). *Proc. roy. Soc. Med.*, **52**, 477.
- Williams, W. O. (1961). *J. Coll. gen. Practit.*, **4**, 181.
- Woodward, T. E., McCrumb, F. R. (Jnr.), Carey, T. N., and Togo, Y. (1960) *Ann. intern. Med.*, **53**, 1130.

Mastitis in the Male. A Rare Complication of Mumps. JOHN S. HAPPEL, M.B., CH.B., D.OBST.R.C.O.G. *Brit. med. J.* 1965. **2**, 1041.

A farmer suffering from typical mumps developed what appeared to be a cellulitis extending from his right axilla to the iliac crest with severe systemic disturbances. There was no response to penicillin and a small ectopic nipple was noticed in the left scapular area. It was considered that the illness was due to mastitis in an embryonic remnant of female secretory breast tissue. The total illness lasted about five weeks.