CASE REPORT

Chest wall myositis in a patient with acute coronary syndrome

Laila Hussein,¹ Harith Al-Rawi²¹Rashid Hospital Trauma Center, Dubai, UAE²St Thomas' Hospital, London, UK

Correspondence to

Dr Laila Hussein,
laila.alawi@yahoo.com

Accepted 23 September 2014

SUMMARY

We describe a case of a 42-year-old man who presented to the emergency department with severe left-sided chest pain and chest tenderness of 1-day duration. The pain was episodic and was aggravated by any chest wall movement. His initial blood tests and ECG were suggestive of acute coronary syndrome (ACS). However, his pattern of pain, lack of response to opiates, raised creatine kinase and signs of pleurisy on chest radiograph raised a suspicion of an alternative diagnosis. The patient showed a dramatic response in pain relief to non-steroidal anti-inflammatory medication. He was suspected to have chest wall myositis with pleural involvement in the form of pleurodynia. His serology test was positive for coxsackie virus antibodies. We will discuss in this case report the pathognomonic features, diagnosis and treatment of a rare infectious condition known as Bornholm disease.

BACKGROUND

Bornholm disease was first described more than 140 years ago. Although there is a good amount of literature published on this condition, it remains a relatively rare disease. Part of its low incidence could be attributed to the fact that it is often missed. As it follows a benign course in the majority of cases and resolves spontaneously, most patients remain undiagnosed. Symptoms of Bornholm disease could be very troubling to patients and it may subject them to unnecessary extensive investigations, and even surgery.

To our knowledge, there are no previous reports of Bornholm's disease occurring in a patient with concomitant coronary artery disease and the link between the two conditions is still unknown.

CASE PRESENTATION

A 42-year-old Pakistani man presented to Rashid Hospital Emergency Department reporting worsening severe chest pain that started gradually the previous night. The patient described the pain as localised around the left nipple extending below the left axilla and to the back. He described it as continuous, sharp and sometimes burning in nature; it fluctuated in severity and was unrelenting. It came in attacks lasting a few minutes with dull pain in between. He gave a pain score of 9/10. The pain worsened in the supine position and was associated with sweating and shortness of breath. He denied any fever, cough, dizziness, palpitations, nausea, diarrhoea or rash and there was no history of any trauma to the chest or abdomen. Review of systems was unremarkable. He also denied any episode of

similar pain in the past. However, the patient mentioned that he had experienced an episode of central chest pain 1 week prior, which was much milder, diffuse and felt more like heaviness. The patient's ECG during his visit to the emergency department at that time revealed ST segment depression in leads III and aVF and a rise in cardiac troponin. He was advised admission and angiography but the patient refused as he was planning to get further workup in his home country. His medical history was significant for poorly controlled diabetes mellitus type II for which he was not taking any treatment. He was also a heavy alcohol consumer and smoked 1 pack of cigarettes per day. He worked in an office in a private family business. The patient had come from Pakistan 1 month prior and there was no history of contact with ill people.

On examination his blood pressure was 140/86 mm Hg, heart rate 88 bpm, respiratory rate 18 breaths/min, temperature 37.1°C and oxygen saturation was 98% on room air. The patient was a middle-aged man of average height and slightly overweight. He looked very anxious and restless and had tears in his eyes. He was holding on to the metal support of the bed and bending forward. He was also continuously getting out of the bed and leaning face forward against the wall, then returning to the bed. On inspection of the site of pain there was no redness, rash or deformity. On palpation, however, the patient had severe tenderness even with the lightest touch over the left side of the chest around the left nipple and below the left axilla. Auscultation of the chest showed good bilateral air entry with no crepitations, wheezing or pleural rub. Heart auscultation revealed normal first and second heart sounds with no murmurs. His abdomen was soft, not distended and there was no organomegaly. Palpation of the abdomen was initially normal, but later significant for tenderness in the epigastric area. Renal angle tenderness was negative. The remainder of the physical examination was unremarkable.

In view of his presentation, ECG and laboratory findings, which are detailed in the investigation section, acute coronary syndrome (ACS) was our differential diagnosis. The patient received 300 mg of aspirin, three doses of 5 mg morphine and three doses of sublingual nitroglycerine. Strangely enough, the patient felt no relief at all. A chest X-ray that was obtained on his previous visit to the emergency department failed to demonstrate any pathology (figure 1). A second chest X-ray was taken and this time it showed minimal pleural thickening over the left diaphragmatic area (figure 2).

CrossMark

To cite: Hussein L, Al-Rawi H. *BMJ Case Rep* Published online: [please include Day Month Year] doi:10.1136/bcr-2014-206664

Figure 1 Chest radiograph taken on the patient's first visit to the hospital demonstrating no pathology.

A senior cardiologist was consulted and a bedside echocardiography was performed, which showed no abnormalities in ventricular wall motion and no signs of heart failure or pericardial effusion. In his opinion the pain was purely musculoskeletal. The patient was given an intramuscular injection of diclofenac with intravenous paracetamol and within an hour had a pain score of 0/10. He was again offered admission and further workup for coronary artery disease but the patient refused admission as he had a flight to catch on the same day.

The next morning the patient returned to the emergency department stating that the same pain had returned 3 h after his discharge and he was unable to travel. Except for a temperature of 37.8°C, his history, physical examination and ECG did not reveal any new findings compared with the day before. However, another chest X-ray was taken and this time it demonstrated a clear patchy pneumonitis in the left lower basal region with minimal left basal pleural effusion ([figure 3](#)). After another diclofenac injection, he was pain free and wanted to be discharged home. He was prescribed oral diclofenac tablets, aspirin and oral Augmentin.

Figure 2 Repeated chest radiograph showing minimal pleural thickening over the left diaphragmatic area.

Figure 3 Chest radiograph taken on the patient's third visit showing a clear patchy pneumonitis in the left lower basal region with minimal left basal pleural effusion.

INVESTIGATIONS

Electrocardiogram (ECG) done on presentation demonstrated sinus rhythm with ST depression of 1 mm in lead III, aVF and Q waves and inverted T waves in lead III and aVF. Repeated ECG the next day did not show any changes from the previous one.

The patient underwent serial blood investigations during his two visits to the emergency department. His full blood count was significant for white cell count (WCC) 12 000/ μ L on his first visit, which had increased to 16 000/ μ L on his second visit with absolute neutrophilia of 12.8000/ μ L. Another alarming result was his rising creatine kinase levels, which are demonstrated in the table below. His urea and electrolytes, including serum calcium, liver function tests, D-dimer and erythrocyte sedimentation rate were all within normal range. As we had suspected at some point the possibility of acute pancreatitis, serum amylase was ordered, which was normal (86 U/L).

As part of his workup for ACS, the following blood results were strongly suggestive of his higher risk for ACS: random glucose 294 mg/dL, triglycerides 421 mg/dL (normal 40–200), cholesterol 193 mg/dL (normal 50–200) and high-density lipoprotein 22 mg/dL (35–120). The pattern of cardiac enzymes rise is demonstrated in the table below. Note that a troponin level between 0.1 and 2.0 is considered as high risk of myocardial damage.

Enzyme	Day 1	Day 7	Day 8
Troponin	0.10	0.13	0.09
CK (normal <190 U/L)	117	191	302
CK-MB (normal <24 U/L)	19	13	15

CK, creatine kinase; CK-MB, CK-myocardial band.

A chest X-ray that was obtained on his previous visit to emergency department failed to demonstrate any pathology ([figure 1](#)). A second chest X-ray was taken and this time it showed minimal pleural thickening over the left diaphragmatic area ([figure 2](#)).

After the patient was discharged the second time and returned for recurrent symptoms, a third chest X-ray was obtained. This

time it demonstrated a clear patchy pneumonitis in the left lower basal region with minimal left basal pleural effusion (figure 3).

DIFFERENTIAL DIAGNOSIS

Initially, the patient was diagnosed and managed as ACS. Although the pain he had described was not typically ischaemic, his risk factors, elevated cardiac troponin and ECG changes were all suggestive of possible non-ST elevation myocardial infarction. Pulmonary embolism was a possibility; however, absence of tachycardia, normal oxygen saturation and normal D-dimer did not support this diagnosis. The severe tenderness the patient felt was more in support of a muscular or cutaneous cause. Diagnosis of Herpetic neuralgia would require vesicular lesions to consequently develop, which was not the case with our patient. His pain description, chest X-ray findings, raised C reactive protein and dramatic response to a non-steroidal anti-inflammatory drug raised the possibility of myalgia with pleural involvement.

OUTCOME AND FOLLOW-UP

Unfortunately we lost contact with the patient after his travel for a period of 4 months. In June he returned to Dubai and informed us that after his discharge from our hospital his symptoms continued for another 3 days and then disappeared. He went back to his home country and was also diagnosed with triple vessel disease and underwent coronary artery bypass surgery. He is now doing well and is symptom free. At this point we tested him for coxsackie IgG antibody, which came back positive.

DISCUSSION

Bornholm disease, today a well-described disorder, was first reported in 1872 by Daae, when he studied a small group of patients with acute pain symptoms in Norway. The initial name given by Daae was Bamble's disease, named after the village it was discovered in. Around 60 years later the name was changed to Bornholm disease by Sylvest who was able to identify 23 patients with similar symptoms while on holiday in Bornholm. It was not until 1950 that a link to coxsackie virus was finally identified by Curnen.¹ Since then, healthcare professionals from around the world have begun to recognise these patterns of symptoms in their patients and 10 of reports have been published, most of which date back to the 1950s and 1960s.

Bornholm disease is a form of viral myalgia, characterised by sudden occurrence of excruciating chest pain (aka pleurodynia), back pain or less commonly abdominal pain. Pain is usually paroxysmal, occurring in attacks that can last for seconds and up to 30 min.¹ During attacks, the pain is severe and intense; in between the attacks, however, patients usually report a constant, dull, pleuritic chest pain. A very important quality of the pain is its direct relationship to sneezing, coughing, laughing and deep breathing, which can cause the patient to look tachypnoeic and even diaphoretic.² The most common location of pain when the thorax is involved is over the lower ribs and is usually unilateral, but it can also occur over the front, back, or substernal area.³ Apart from pain, patients can present with a wide range of signs and symptoms. Listed according to descending order of frequency, patients could manifest with fever (97%), pharyngitis (85%), headaches (50%), gastrointestinal disturbances (50%), chest wall tenderness (25%), otitis (25%), dermatitis (25%) and testicular pain (10%).⁴

Coxsackie virus infections are more common in summer and may cause epidemics. Children are more commonly affected

with coxsackie and are more likely to have involvement of the upper abdominal wall, whereas adults more often develop pleurodynia. Infants with Bornholm disease may mimic intussusception, whereas older children and adolescents may present with concern for appendicitis or perforated peptic ulcer. Spread is through direct contact with an infected person. Following an incubation period of 1–4 days in the gastrointestinal tract it then spreads through the haematogenous route.²

Diagnosis of Bornholm disease is made on clinical grounds. However, a few laboratory findings can aid in the diagnosis. A full blood count could show a picture of leucopenia or leucocytosis with or without elevated band cells,⁵ but a normal WCC is not uncommon. There are also a number of reports of elevated erythrocyte sedimentation and creatine kinase. IgM and IgG to coxsackie virus are both equally helpful, however, IgA has the highest specificity.¹ Negative serology does not rule out the disease. In one study, out of 259 patients with diagnosed disease, only 70 had positive IgM antibodies.⁴ The most specific test, but not sensitive, is virus isolation from stool, pleural fluid or the pharynx for up to 2 weeks after resolution of symptoms. The most common isolated variant was found to be coxsackie virus B1-B6, and less commonly A9, echovirus.⁴ As our patient refused to be admitted and undergo further testing, we were unable to isolate the virus and identify the strain he had. He was only tested for IgG antibodies, which were positive. Chest radiography, abdominal ultrasound and CT scan are usually normal.¹ However, there are several reports of positive lung infiltrates with or without pleural effusion as was the case with our patient. Huang *et al*⁵ reported that 45% of patients with Bornholm disease had positive chest radiological findings.

The course of the disease is usually short, lasting 5–7 days but return to normal health may be gradual after a period of weakness and fatigue. There are also reports of pain lasting up to 45 days. In uncomplicated cases treatment is supportive with only good analgesia coverage. Non-steroidal anti-inflammatory drugs usually have satisfactory results. Our patient showed dramatic response to intramuscular diclofenac injections. In more resistant cases, there are successful reports of infiltration of the intercostal nerves with 2% xylocain.⁶ No deaths are reported as a direct result of pleurodynia, but there are a number of case reports of serious complications such as myocarditis, pericarditis and encephalitis. Most of these complications occur in extremes of age or immunocompromised patients. Tang *et al*⁷ described a case of neonatal encephalitis in an infected pregnant patient. Although there are reports of myocarditis and pericarditis, to our

Learning points

- ▶ A hallmark of Bornholm disease is paroxysmal chest or abdominal pain that is aggravated by chest wall movement.
- ▶ Symptoms can easily be mistaken for acute coronary syndrome or acute abdomen.
- ▶ Although no test is highly sensitive, serology and viral isolation from stool remain the most specific tests and should be ordered on all suspected cases.
- ▶ Pain that responds to non-steroidal anti-inflammatory drugs is very likely to be muscular or pleuritic in origin.
- ▶ Bornholm disease can occur in epidemics, and physicians should keep this diagnosis in mind when family members or students from the same school present with similar symptoms.

knowledge there are no previous cases in the literature of Bornholm disease in a patient with underlying myocardial ischaemia. Although we do not have detailed reports of the coronary angiography that our patient underwent in his home country, while in our emergency department he had clear ST segment depression on his ECG and a documented elevation of troponin. Whether ACS may be a risk factor for coxsackie virus infection or there is a direct link between the two conditions is unclear.

Contributors LH diagnosed and managed the patient mentioned in the case report. She also wrote the case report and did the literature review. She is guarantor. HA-R was the attending consultant when the patient presented to the emergency department. He reviewed the case report and made the necessary corrections to language and gave his suggestions.

Competing interests None.

Patient consent Obtained.

Provenance and peer review Not commissioned; externally peer reviewed.

REFERENCES

- 1 Ivan Y, Kelly BP. Bornholm disease case report and a review of the literature. *Infect Dis Clin Pract* 2013;00:00–00.
- 2 Hopkin JHS. Bornholm disease. *Br Med J* 1950;1:1230–2.
- 3 Kliegman RM, Stanton B, St. Geme J, et al. Non-polio enteroviruses. *Nelson textbook of pediatrics*. 19th ed. Philadelphia, PA:Elsevier Saunders, Inc. 2011:1088–94.
- 4 Schmidt NJ, Magoffin RL, Lennette EH. Association of Group B coxsackieviruses with cases of pericarditis, myocarditis, or pleurodynia by demonstration of immunoglobulin M antibody. *Infect Immun* 1973;8:341–8.
- 5 Huang WT, Lee PI, Chang LY, et al. Epidemic pleurodynia caused by coxsackievirus B3 at a medical center in northern Taiwan. *J Microbiol Immunol Infect* 2010;43:515–18.
- 6 Georgios T, Dimos K. Bornholm disease—a pediatric clinical entity that can alert a thoracic surgeon. *J Paediatr Child Health* 2011;47:240–4.
- 7 Tang JW, Bendig JW, Ossueta I. Vertical transmission of human echovirus 11 at the time of Bornholm disease in late pregnancy. *Pediatr Infect Dis J* 2005;24:88–9.

Copyright 2014 BMJ Publishing Group. All rights reserved. For permission to reuse any of this content visit <http://group.bmj.com/group/rights-licensing/permissions>.
BMJ Case Report Fellows may re-use this article for personal use and teaching without any further permission.

Become a Fellow of BMJ Case Reports today and you can:

- ▶ Submit as many cases as you like
- ▶ Enjoy fast sympathetic peer review and rapid publication of accepted articles
- ▶ Access all the published articles
- ▶ Re-use any of the published material for personal use and teaching without further permission

For information on Institutional Fellowships contact consortiasales@bmjgroup.com

Visit casereports.bmj.com for more articles like this and to become a Fellow