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Case report

Unusual cause of chest pain, Bornholm disease, a forgotten entity; case report and review of literature

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ABSTRACT

Chest pain is a common symptom culminating in hospital admissions and specialist referrals. Although cardiac work up is pursued in most of the cases, cardiac etiology is found to be the culprit in minority of the cases. Acute chest pain is a clinical syndrome that may be caused by almost any condition affecting the thorax, abdomen, or internal organs. On occasions this extensive and expensive diagnostic work up can be avoided with awareness of commoner and non-lethal reasons. We report a case of a woman with Bornholm disease secondary to Coxsackievirus B5 (CB5) infection and supplementary review of literature till date.

1. Introduction

Chest pain is a common symptom culminating in hospital admissions and specialist referrals. Although cardiac work up is pursued in most of the cases, cardiac etiology is found to be the culprit in minority of the cases [1–5]. Data from epidemiologic studies have indicated that the overall proportion of non-cardiac chest pain (NCCP) among patients with chest pain is reported between 20 and 40%.This fraction is robust and internationally similar in different countries, such as Germany Europe, USA, China, or Australia [6]. Acute chest pain is a clinical syndrome that may be caused by almost any condition affecting the thorax, abdomen, or internal organs. When a patient presents with chest pain in the emergency room, the first priority is to consider life-threatening causes such as acute coronary syndromes, aortic dissection, pulmonary embolism (PE), ruptured aortic aneurysm, and tension pneumothorax [7]. The diagnostic work up for these etiologies bear a rightful demand on time of the emergency room physicians and other resources. On occasions this extensive and expensive diagnostic work up can be avoided with awareness of commoner and non-lethal reasons. Absence of knowledge of this entity often confuses clinicians to pursue the alternative diagnoses such as acute abdomen and acute chest syndromes including pleurisy and coronary syndromes. We report a case of a woman with Bornholm disease secondary to Coxsackievirus B5 (CB5) infection and supplementary review of literature till date.

2. Case

A 68-year-old female was admitted to the hospital with acute onset and progressively worsening sharp epigastric and left sided “gripping” chest pain of severe intensity without any radiation or change in character with position, two days prior to the presentation. The onset was without any trauma. Her past medical history was remarkable for hypertension, hyperlipidemia and hypothyroidism. Her medications included aspirin, amlodipine, lovastatin and levothyroxine. Patient described her pain as pleuritic in nature with exacerbation on taking a deep breath and no position relieved the pain. Patient denied any recent travel. There were no family members with similar symptomatology. On examination, patient was in significant discomfort and clutching pain in the left side of chest and epigastrium. She had normal blood pressure but was mildly tachycardic with a heart rate of 105 beats per minute. She had a low grade temperature of 99.9 degree Fahrenheit. Her oxygen saturation was 97% without any supplemental oxygen. The pain was not reproducible on palpation and there was no visible rash. Upon auscultation of the chest there was appreciable pleural rub on the left, and the right lung examination was unremarkable. Rest of her systemic examination was unremarkable. Patient was given appropriate

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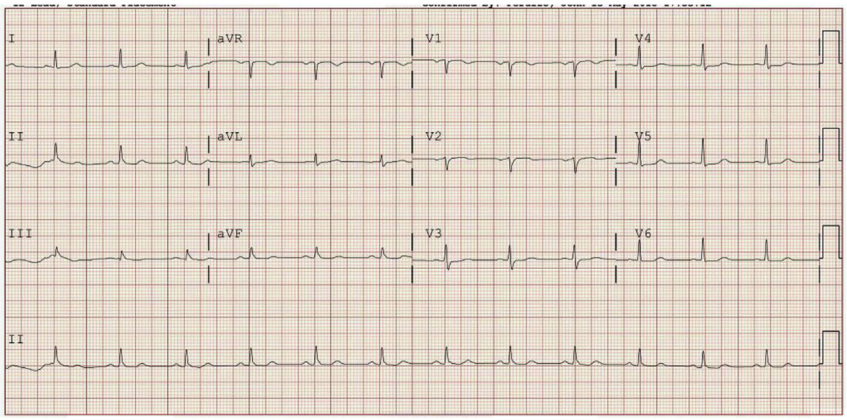

Fig. 1. EKG showing normal sinus rhythm with no ST-T wave changes suggestive of any ischemia.

analgesia.

Laboratory data showed normal white blood cell count of 9.4 × 1000/μL (3.4–11.0 × 1000/μL) with 54% neutrophils, along with normal hemoglobin and hematocrit. Serum chemistry was normal with creatinine of 0.75 mg/dL (0.5–1.5 mg/dL) and normal liver function tests. Due to concerns of chest pain and acute coronary syndrome her cardiac biomarkers were checked and troponin T was found to be normal (less than 0.030 ng/mL) on three occasions, each 6 hours apart. Creatine kinase (CK) was normal 122 U/L (24–173 U/L). MB component of CK was found to be normal as well at 3.7 ng/mL (0.0–5.3 ng/mL). EKG done for the patient to rule out any ongoing cardiac ischemia was normal (Fig. 1) without any st wave changes.

Chest x-ray done showed small bilateral effusions (Fig. 2).

Due to unclear diagnosis of pleuritic chest pain at this time and sinus tachycardia, patient underwent computed tomography (CT) of the chest to rule out pulmonary embolism and was found to be negative for any embolic event or pulmonary infarction, aortic abnormality and gastro-esophageal abnormality. In addition there was no muscular edema noted on the scan. After ruling out the most ominous causes of chest pain, patient was still symptomatic. Now suspicion was high for rare causes of chest pain. Wider net was now casted for vasculitic disorders and rare infections causing similar symptomatology. Erythrocyte sedimentation rate and C-reactive protein were elevated to 65 mm/hr (2–15 mm/hr) and 17.2 mm/hr (0.0–4.9 mg/L) respectively. Her complement levels for C3 were 161 mg/dL (82–167 mg/dL) and for C4 were 16 mg/dL (14–44 mg/dL). Her C-ANCA and p-ANCA panel was negative. Antinuclear antibodies were negative. Further serological testing was done and she was found to have negative serology for coxsackievirus A types 7, 9, 16 and 24. Finally serology returned positive for coxsackievirus B type 5 (CB5) by complement fixation reaction (CFR, Virion/Serion, Clindia Benelux b.v., Almere, the Netherlands) and negative for coxsackievirus B types 1, 2, 3, 4 and 6.

Patient was now continued to be treated as epidemic pleurodynia or Bornholm disease (caused by CB5) with symptom control using non-steroidal anti-inflammatory drugs and intravenous fluids. Patient's pain improved significantly over the next 48 hours with resolution of fever and tachycardia. Patient was discharged home on the fourth day with complete resolution of symptoms. At 2 weeks follow up, patient was completely chest pain free. Repeated chest x-ray at this time showed normal lung fields with clearance of bilateral small pleural effusions (Fig. 3).

3. Discussion

First described in Daae and Homann in Norway, disease has been known with many forenames such as Bornholm (City in Denmark) disease, Devil's grippe, epidemic myalgia and epidemic pleurodynia [8]. However in the early years there was no identifiable pathogen associated with the disease. Since then there have been glaring advances in microbiological and virological sciences. It was not until 1949

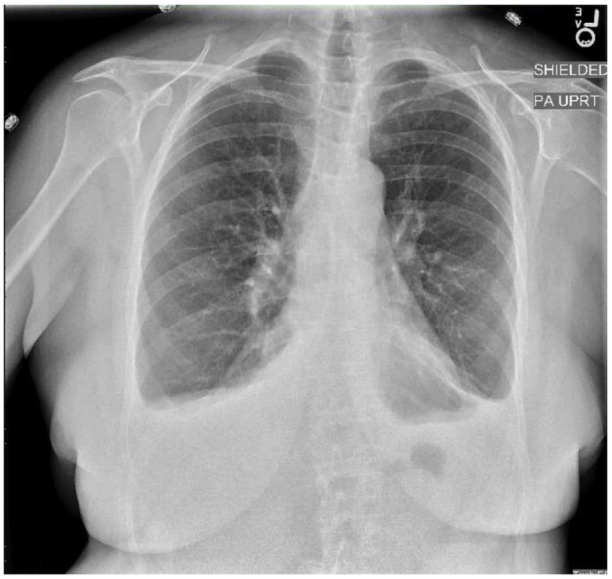

Fig. 2. Chest radiograph showing small bilateral pleural effusions with no airspace disease.

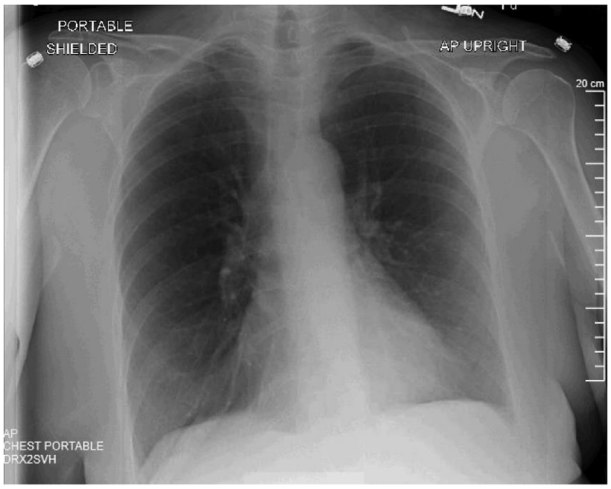

Fig. 3. Chest radiograph repeated after 2 weeks showing complete resolution of pleural effusions.