

CASE NOTE

Large pleural effusion necessitates chest tube drainage in a patient with Kawasaki disease

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Abstract: An 11-month-old Turkish boy was hospitalised with clinical and roentgen graphic evidence of large pleural effusion on the third day of fever and misdiagnosed as parapneumonic effusion. Due to worsening respiratory distress chest tube drainage was performed. Four days later the classic signs of Kawasaki disease appeared. His clinical condition improved gradually and fever subsided after intravenous gammaglobulin and aspirin treatment. A mild transient dilatation of the right coronary artery was seen and returned to the normal diameter within a few weeks. To our knowledge, large pleural effusion in a case of Kawasaki disease, in which chest tube drainage was needed, has not been reported. We describe here a patient with complete Kawasaki disease whose initial presentation mimicked a parapneumonic effusion.

Key words: complication; Kawasaki disease; pleural effusion.

Kawasaki disease (KD) is an acute febrile exanthematous illness of childhood characterised by multi-organ involvement. The diagnosis of KD is based on clinical features and supporting laboratory, and echocardiographic findings. There have been a few reports of significant pleural effusion causing pulmonary dysfunction associated with KD.¹⁻⁵ Here we describe a male infant who presented with severe respiratory distress due to large pleural effusion and subsequently developed classical features of KD.

To our knowledge, this is the first report of KD in which pleural effusion preceded the classical features of KD and chest tube drainage was performed due to worsening respiratory distress.

Case Report

A previously healthy 11-month-old Turkish boy presented with fever and irritability. Examination revealed a fever of 39°C and pharyngeal erythema. Treatment with amoxicillin was given. On the third day of fever, he developed dyspnea and was admitted to the local hospital. Physical examination revealed a body

temperature of 39°C, respiratory rate of 60/min and lessened breath sounds in the right lung. Abdominal examination and cardiac auscultation were unremarkable. Laboratory studies showed an increased C-reactive protein (218 mg/L), leukocytosis ($32.8 \times 10^3/\text{mL}$), mild anaemia (10.4 g/dL), increased leukocytes in urine sediment, and normal liver function test. Chest radiograph and chest computed tomography revealed the right lung to be markedly compressed by the huge pleural effusion and the mediastinum being shifted to the left side (Fig. 1). As a remedy, antibiotic therapy with ceftriaxone and vancomycin was started with a diagnosis of parapneumonic effusion. Thoracentesis was performed due to increasing dyspnea, lowering oxygen saturation and moderate respiratory acidosis on the second day of admission (day 4) and 650 mL pleural fluid was drained within 5 days. Pleural fluid was transudate with a white blood cell count of $1.3 \times 10^3/\text{mL}$ (40% neutrophils, 60% lymphocytes), pleural fluid/serum LDH ratio = 0.45, pleural fluid/serum protein ratio = 0.44. No pathogen was found in the culture of the pleural effusion. Culture and polymerase chain reaction analysis for tubercle bacilli were found to be negative. The patient developed bilateral conjunctival injection, disseminated erythematous macules of the trunk and extremities, perineal erythema, dorsal oedema of the hands and feet, strawberry tongue and cheilitis on day 7. The diagnosis of KD was made in the presence of four of the five classical clinical criteria, in addition to fever duration of more than 5 days. The patient was seen by one of the authors (AD) and the echocardiographic examination did not revealed any abnormality. He received a single dose of 2 g/kg gammaglobulin intravenously (IVIg) and high dose aspirin (80 mg/kg/day) was initiated, while the antibiotics were stopped. Fever subsided on the next day and his clinical condition gradually improved. During this time, the thrombocyte count rose to $788 \times 10^3/\text{mL}$. Desquamation of the hands and feet were noticed on day 12. On the fourteenth day of the disease, the patient's fever rose again and he was referred to our paediatric cardiology department on day 15. Upon

Key Points

- 1 Misdiagnosis can be made if unusual complications of Kawasaki disease (KD) precede the classical signs.
- 2 Large pleural effusion can be the presenting manifestation of KD.
- 3 Chest tube drainage may be required due to respiratory failure in KD.

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Fig. 1 Computed tomography on day 3 of the illness revealed the right lung to be markedly compressed due to huge pleural effusion and the mediastinum dramatically shifted to the left side.

admission, physical examination showed the infant was febrile (38.7°C), tachypneic (45/min) and tachycardic (135/min). Desquamation over the perineal region was noticed. The laboratory findings of his condition at our hospital showed an erythrocyte sedimentation rate of 90 mm/h, a C-reactive protein level of 60 mg/L, a white blood count of $19.8 \times 10^3/\text{mL}$, thrombocytosis of $734 \times 10^3/\text{mL}$ and a hemoglobin level of 8 g/dL. Serology for viruses (EBV, CMV, parvovirus B19, HIV and hepatitis A, B), IgM antibody to mycoplasma pneumonia and anti-streptolysin O titres were negative. Complement 3 and 4, antinuclear antibody and rheumatoid factor were normal. An echocardiogram revealed the presence of a dilatation (4 mm) and perivascular echo-brightness of the right coronary artery with normal myocardial contractility. A repeat chest computed tomography was unremarkable. Slit-lamp examination showed numerous, minute, non-pigmented keratic precipitates in both of the patient's eyes. No vitreous or fundal abnormalities were observed. A second dose of IVIG (2 g/kg) infused on day 16. The fever finally subsided and he was discharged from hospital on day 20 with low-dose aspirin. Follow-up after 2 months showed a resolution of anterior uveitis, normal erythrocyte sedimentation rate and right coronary artery.

Discussion

Kawasaki disease is an acute vasculitis of unknown aetiology with varied clinical manifestations. Despite numerous efforts, there is still no diagnostic test available for KD, and the diagnosis is based on clinical criteria after the exclusion of other diseases presenting with high and persistent fever. Cardiac involvement is considered to be the most important complication of KD. From the echocardiographic studies, approximately 40% of the patients with KD revealed coronary artery dilatation in an acute stage.⁶ We have described an infant with KD manifesting initially as large pleural effusion, and who required

chest tube drainage due to respiratory distress. Fortunately, classical features of KD before the development of coronary artery abnormalities appeared soon after hospitalisation and treatment with IVIG was initiated. Early IVIG therapy reduces the risk of giant coronary aneurysm. Hence, early diagnosis and treatment are crucial.

In a series of 129 patients with KD, 19 (14.7%) had chest radiograph abnormalities. Of the 19 cases, three cases (15.8%) had pleural effusion.⁷ Significant pulmonary involvement is unusual and the prognosis of pulmonary complications in KD is generally good. A significant amount of pleural effusion associated with KD has rarely been reported in literature.¹⁻⁵ The reported cases were treated with IVIG or methylprednisolone. In our case, large pleural effusion was the presenting manifestation of KD and was misdiagnosed as parapneumonic effusion. As respiratory distress had worsened, chest tube drainage was performed despite the fluid being transudate. We found no other reports in the English literature of KD and large pleural effusion preceding classical signs of KD and in which chest tube drainage was required due to respiratory failure. IVIG was given immediately upon diagnosis of complete KD. As a result, the pleural effusion and other clinical features had dramatically improved. A repeat dose was needed due to the recurrence of fever. A mild dilatation and perivascular echo-brightness of the right artery were seen in our patient and was later returned to the normal diameter within a few weeks.

In conclusion, this patient presented in an unusual manner. We would like to draw attention to the fact that as with many other vasculitic diseases, KD can sometimes predominantly involve the respiratory system. In addition, we have shown that misdiagnosis can be made should unusual complications of KD precede the classical mucocutaneous signs, even in cases with complete KD. In such cases, physicians should be aware of the various clinical presentations of KD.

References

- Dahlem PG, von Rosenstiel IA, Lam J, Kuijpers TW. Pulse methylprednisolone therapy for impending cardiac tamponade in immunoglobulin-resistant Kawasaki disease. *Intensive Care Med.* 1999; **25**: 1137-9.
- Voynow JA, Schanberg L, Sporn T, Kredich D. Pulmonary complications associated with Kawasaki Pediatric disease. *J. Pediatr.* 2002; **140**: 786-7.
- Sittiwangkul R, Pongprot Y. Large pleural effusion: an unusual manifestation of Kawasaki disease. *Clin. Pediatr.* 2004; **43**: 389-91.
- Hamada H, Terai M, Honda T, Kohno Y. Marked pleural and pericardial effusion with elevated Vascular Endothelial Growth Factor production: an uncommon complication of Kawasaki disease. *Pediatr. Int.* 2005; **47**: 112-14.
- D'Souza S, Khubchandani RP, Shetty AK. Kawasaki disease presenting with hemorrhagic pleural effusion. *J. Trop. Pediatr.* 2005; (Epub ahead of print).
- Kato H. Cardiovascular complications in Kawasaki disease: coronary artery lumen and long-term consequences. *Prog. Pediatr. Cardiol.* 2004; **19**: 137-45.
- Umezawa T, Saji T, Matsuo N, Odagiri K. Chest x-ray findings in the acute phase of Kawasaki disease. *Pediatr. Radiol.* 1989; **20**: 48-51.