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Risk factors for retropharyngeal cellulitis in Kawasaki disease

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ABSTRACT

Objective: Kawasaki disease (KD) is an acute multisystemic vasculitis of unknown etiology that occurs in infants and children. Retropharyngeal cellulitis has been reported as a rare manifestation of KD. This study investigated the frequency and characteristics of patients with KD manifesting as retropharyngeal soft-tissue swelling.
Methods: We retrospectively reviewed 277 patients, with a mean age of 1 year and an age range of 7 months to 12 years, in whom KD had been diagnosed between 2005 and 2011.
Results: In 10 patients (3.6%), contrast-enhanced computed tomography (CECT) showed low-density lesions without ring enhancement in the retropharyngeal spaces. These patients presented initially with fever and cervical lymphadenopathy, and were initially treated by their pediatricians for suppurative lymphadenitis (seven patients) or retropharyngeal abscess (three patients). KD was finally diagnosed either after antibiotics had been ineffective or when other symptoms characteristic of KD emerged.
Conclusion: Low-density lesions in the retropharyngeal space were identified by CECT in 3.6% of the KD patients. Early diagnosis of KD is essential because coronary artery lesions develop in 50% of untreated patients. If a child presents with fever, cervical lymphadenopathy, and swelling of the retropharyngeal space, KD should be included in the differential diagnoses.

1. Introduction

Kawasaki disease (KD) is an acute multisystemic vasculitis of unknown etiology that occurs in infants and children. It is characterized by fever, conjunctivitis, changes in the oral mucous membranes, changes in the peripheral extremities, skin rash, and cervical lymphadenopathy [1,2]. KD is sometimes difficult to diagnose as there is no specific laboratory test for this disease, and additionally, symptoms that match the diagnostic criteria for KD might not be present simultaneously, but rather can appear in sequence. A delay in diagnosing KD can be serious, resulting in the development of coronary artery aneurysms, which could lead to myocardial infarction, ischemic heart disease, or sudden death [3]. Some KD cases have initially presented as a retropharyngeal abscesses or cellulitis [4–17]. Retropharyngeal abscess also mainly affect children, causing fever, cervical lymphadenopathy, leukocytosis, and elevated serum C-reactive protein (CRP). As both KD and retropharyngeal abscess can be fatal if an appropriate treatment is not provided, differentiating these diseases is crucial. Nevertheless, the pathophysiology and the incidence of retropharyngeal lesions in KD have rarely been reported. We describe a 7-year-old female who was originally diagnosed with a retropharyngeal cellulitis, but was subsequently treated successfully for KD, which prompted us to review similar cases to investigate the incidence of KD mimicking retropharyngeal abscess or cellulitis.

2. Materials and methods

2.1. Case study

A previously healthy 7-year-old girl who had been suffering from a fever of up to 39 °C for 2 days and cervicodynia consulted pediatricians at Kobe City Medical Center General Hospital (Kobe, Japan). She had been treated with cefditoren pivoxil for 2 days. Physical examination revealed tender and enlarged left cervical lymph nodes, torticollis, and trismus; however, no other symptoms of KD (skin rash, conjunctivitis, lip fissuring, or strawberry tongue) were observed. The patient was unable to shake her head from side to side because of the severity of the cervicodynia. The

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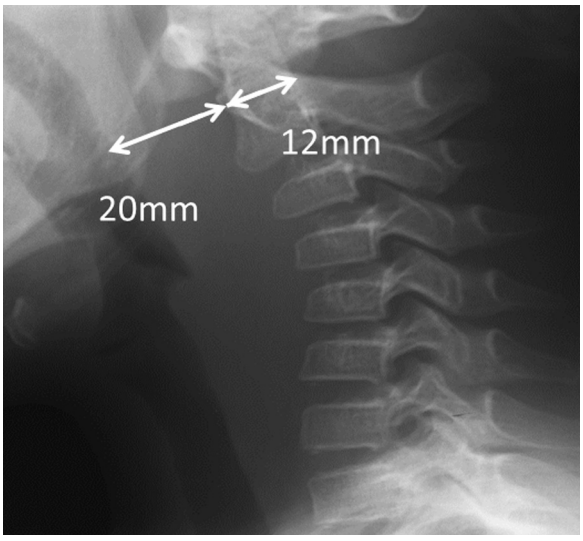

Fig. 1. Lateral neck radiograph showing marked retropharyngeal soft-tissue swelling, which was 1.7 times as wide as the C2 vertebral body.

pediatricians suspected the presence of a retropharyngeal abscess, and performed a lateral neck radiography and neck contrast-enhanced computed tomography (CECT) to investigate.

The patient had the following initial laboratory test results: white blood cell count (WBC), 28,600/μL; hematocrit, 38.7%; platelet count, 35.0×10^4/μL; CRP level, 9.5 mg/dL; sodium level, 135 mequiv./L; albumin level, 4.7 mg/dL; aspartate aminotransferase (AST) level, 16 IU/L; and alanine aminotransferase (ALT) level, 6 IU/L.

Lateral neck radiography showed retropharyngeal soft-tissue swelling (Fig. 1). A CT scan showed a low-density lesion in the retropharyngeal space without ring enhancement (Fig. 2), with multiple cervical lymph nodes gathering into one palpable mass with a non-enhancing center. Retropharyngeal cellulitis was suspected at this point. Pharyngeal endoscopy showed no sign of topical inflammation of the pharynx and so surgery was not considered appropriate for this patient.

The patient was treated with the antibiotic ceftriaxone; however, her symptoms did not improve. On day 6, the patient developed a skin rash, lip fissuring, and strawberry tongue, and the diagnosis of KD was made. She was given 2 g/kg intravenous immunoglobulin immediately, and the fever subsided and the skin rash, lip fissuring, and strawberry tongue disappeared in 2 days. The cervical lymph nodes also gradually reduced in size, and the retropharyngeal soft-tissue swelling was no longer visible by lateral neck radiography on day 10, when the patient was discharged from hospital. Repeated echocardiograms revealed no coronary artery abnormalities.

2.2. Retrospective cohort study

We retrospectively reviewed the medical records of 277 patients who were diagnosed with KD and treated by pediatricians at Kobe City Medical Center General Hospital between January 2005 and December 2011. The patients were aged from 0 to 12 years, with a mean age of 1 year, and comprised 171 (61.7%) boys and 106 (38.3%) girls.

Patients with KD who had cervical CECT were selected from their medical charts, and those with low-density lesions in the retropharyngeal space were identified. We compared patients with and without low-density lesions in the retropharyngeal space using seven criteria: age, gender, presence of cervical lymphadenopathy, WBC, CRP levels, time from onset of fever to diagnosis of KD, and coronary artery abnormalities. WBC and CRP levels were investigated by initial laboratory data, and coronary artery abnormalities were evaluated by repeated echocardiograms after diagnosis of KD.

2.3. Statistical analysis

The Mann–Whitney *U* test was used to compare age, WBC, CRP levels and length of illness before diagnosis of KD between patients with and without low-density lesions in the retropharyngeal space. The chi-square test was used to compare gender, cervical lymphadenopathy, and coronary artery abnormalities between the two groups. Values of $P < 0.05$ were considered statistically significant.

3. Results

Of the 277 patients diagnosed with KD, 13 (4.7%, median age, 5 years) had CECT for suspected retropharyngeal abscess or cervical lymphadenopathy. No other patients had clinical symptoms suggestive of cellulitis. A low-density lesion in the retropharyngeal space was identified in 10 patients (3.6%; Table 1). These 10 patients all initially experienced fever, cervicodynia, and cervical lymphadenopathy. Their pediatricians had initially treated seven

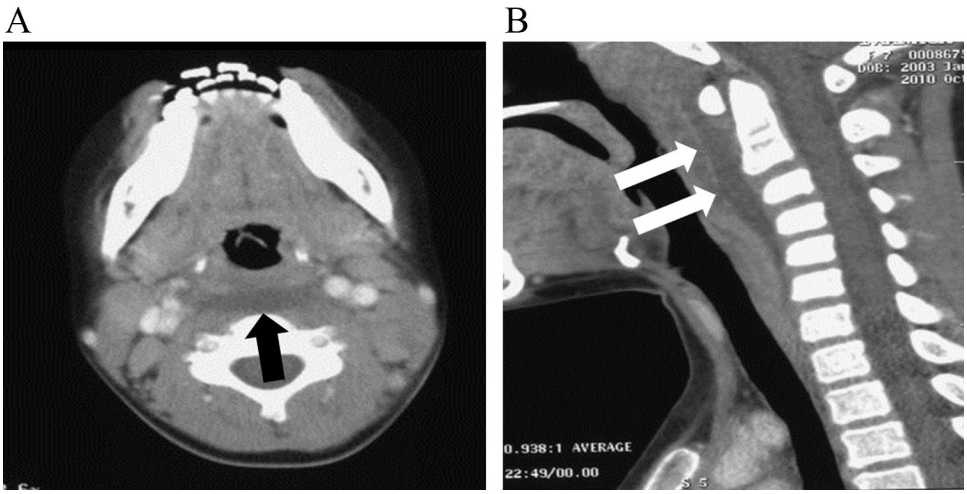

Fig. 2. Enhanced neck CTs showing a low-density lesion (arrows in a and b) in the retropharyngeal space. (a) Axial view, showing no significant ring enhancement. (b) Sagittal view.