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Case report

Retropharyngeal abscess: An unusual presentation of Kawasaki disease. Case report and review of the literature

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ABSTRACT

We report the case of a patient with Kawasaki disease whose initial presentation mimicked a retropharyngeal abscess and review the literature of this topic (16 cases reported). Fever and deep neck infection like symptoms were the only clinical findings at admission in 87.5% children. All children had a neck CT scan performed showing findings suggestive of retropharyngeal abscess. All children were started antibiotic therapy without clinical improvement and 31% of patients underwent unproductive surgical drainage of the retropharyngeal space. Otolaryngologist should be aware of atypical presentation of Kawasaki disease presentation mimicking retropharyngeal abscess. Early diagnosis is pivotal for preventing cardiac complications and avoiding the risk associated to unnecessary surgical intervention.

1. Background

Kawasaki disease (KD) is an acute self-limiting vasculitis of childhood of unknown etiology, which affects small- and medium-sized arteries. Early diagnosis of KD is pivotal for a timely treatment with intravenous immunoglobulin to reduce the potentially life-threatening cardiological complications [1]. Diagnosis is based on well defined clinical criteria which include: fever ≥5 days, cheilitis and oral mucositis, polymorphous skin rash, cervical lymphadenopathy and nonexudative conjunctivitis. Onset of this disease may be subtle and present with febrile symptoms mimicking other conditions such as acute neurological or otolaryngological manifestations [2]. A few cases of KD mimicking retropharyngeal abscess have been reported in the literature [3–12]. Early recognition of this uncommon KD presentation by the otolaryngologists is essential for a proper management of these children.

Here we report a case of a child with final diagnosis of KD who presented as a retropharyngeal abscess and review the literature on this topic.

2. Case report

A previously healthy 4 year old girl presented to our emergency department with a 24 h history of fever (up to 39 °C), right neck swelling and pain associated with restricted neck movements. At presentation she was still under antibiotic treatment with amoxicillin for an upper respiratory tract infection and left acute otitis media diagnosed 6 days before. On admission she was febrile and ill-appearing. On physical examination restriction in neck extension movements, right firm enlargement of the jugular chain cervical lymph nodes and hyperemic throat were observed. The remaining of the physical examination was normal. Laboratory evaluation revealed a white blood cell count (WBC) of 40.480/mm^3 with 86.8% neutrophils, a hemoglobin concentration of 10.7 g/dL and a platelet count of 576.000/mm^3; C reactive protein (CRP) was increased to 6.5 mg/dL and erythrocyte sedimentation rate (ESR) resulted 110 mm/h. Blood culture and pharyngeal swab were undertaken.

MRI was performed revealing considerable bilateral cervical lymphadenopathies, more extensive in the right side, with the largest node measuring about 3 cm; in the right retrotonsillar and retropharyngeal spaces, the normal fat disappeared and a hyperintense area on fat-suppressed (FS) T2 sequence was

Abbreviations: KD, Kawasaki disease; WBC, white blood cell count; ESR, erythrocyte sedimentation rate; CRP, C reactive protein; MRI, magnetic resonance imaging; CT, computed tomography; FS, fat-suppressed; IVIG, intravenous immunoglobulin.

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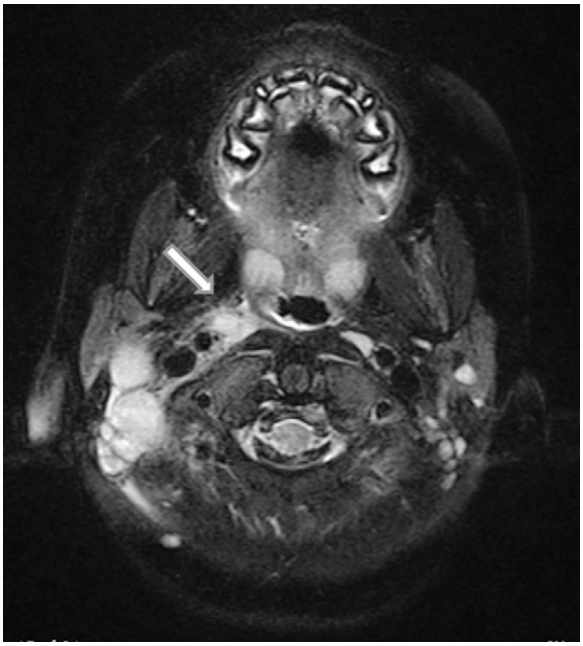

Fig. 1. MRI T2 sequence image: Axial T2 FS sequence shows enlarged high signal reactive cervical adenopathy, mainly in the right posterior cervical space. A hyperintense area is observable in retropharyngeal and retrotonsillar right space, with mildly bulging to pharyngeal wall (*arrow*).

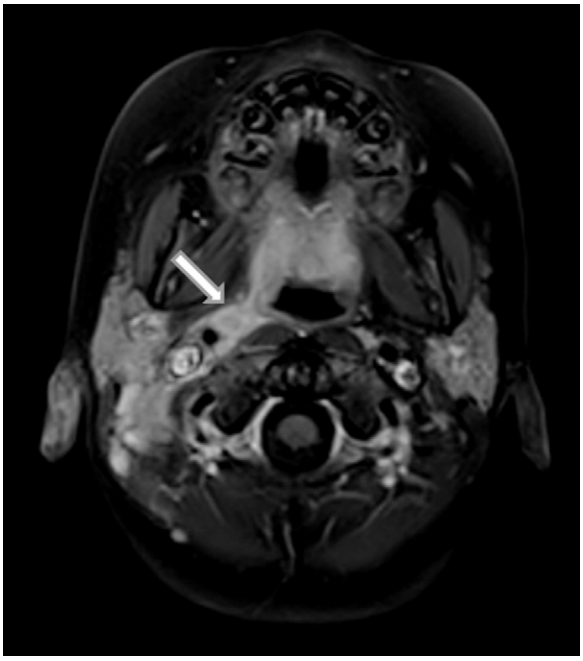

Fig. 2. MRI T1 sequence image: Axial T1 sequence after gadolium show a diffuse enhacement in the retropharyngeal–retrotonsillar space and a nodular area about 1 cm with just mildly enhancing rim, suggestive of an abscess (*arrow*).

observed (Fig. 1). On T1 FS sequence a suppurative node, with mild enhancing rim and necrosis inside, was noted after gadolium contrast injection, suggestive of a retropharyngeal abscess. The inflammatory process spread to adjacent spaces in the same side, with a mildly extension to the prevertebral space; no mediastinal extension was observed (Fig. 2).

The patient received intravenous antibiotic therapy with ceftriaxone and clindamicin without any clinical and laboratory improvement in the following four days. Microbiological investigations were ultimately negative.

On hospital day 5, the patient developed a bilateral non purulent conjunctivitis, a strawberry tongue with fissuring of lips and stomatitis consistent with Kawasaki disease. An echocardiogram was performed and showed perivascular coronary artery brightening without other abnormalities. The electrocardiogram was normal. At this time laboratory investigations revealed WBC 28.740/mm^3 (with neutrophils 87% and lymphocytes 11%); hemoglobin 9.5 g/dL; platelet count 513.000/mm^3; CRP 7 mg/dL, ESR 120 mm/h, sodium 139 m equiv./L, AST 21 U/L, ALT 61 U/L, total protein count 5.9 g/dL, Albuminemia 3.3 g/dL.

Treatment with intravenous immunoglobulin (IVIG) and high dose aspirin was immediately started and was followed by rapid and marked clinical improvement. Within 24 h the child became steadily afebrile and surgical drainage was not performed.

She was discharged on hospital day 10 with complete resolution of neck stiffness and a reduction of lymphoadenopathy. Her WBC and CRP at discharge decreased to 5.860/mm^3 and 1.3 mg/dL respectively.

An echocardiogram performed one week after discharge revealed no evidence of cardiac abnormalities.

3. Review of literature

A pubmed search of English literature was carried out using the following key words (Kawasaki disease OR Mucocutaneous lymph node syndrome) AND retropharyngeal abscess. Reference search of the retrieved articles was also performed.

Nine papers reporting 15 cases of KD mimicking a retropharyngeal abscess were found through the pubmed search, while one more case was retrieved through reference hand search, for a total of 16 cases (Table 1).

Patients were predominantly male (11/16, 69%), with an age range between 10 months and 9 years, with only 1 case reported in Europe. Fever and deep neck infection like symptoms were the only clinical findings at admission in 14 patients (87.5%) [4–12]. The same number of children had a fever duration $\leq$5 days at presentation [3–7,9,10,12] and only 2 patients presented a polymorphous rash [3,4].

All children had a neck CT scan performed showing findings suggestive for retropharyngeal abscess.

Intravenous antibiotics were initially administered to all patients with no response during the following hospitalization days. Five children (31%) [3,4,6,8,9] underwent surgical exploration of the retropharyngeal space. Neither tissue fluctuance nor purulent fluid collection was found in any of them.

In all patients KD diagnosis was suspected based on additional clinical findings, which presented later on during hospitalization. Five children (31%) [3,4,6,12] were finally diagnosed with incomplete KD, four of them [3,6,12] fulfilling 3 criteria besides fever duration, and one patient presenting only 2 criteria [4]. Twelve patients [3–12] of the fifteen for whom the data was reported (80%) received a diagnosis of KD, after 7 or more days since onset of fever and 4 (26.7%) [4,6,8,11] after 10 or more days. Eight patients (50%) [4–9,12] showed abnormal findings on echocardiography, including: pericardial effusion (3 cases) [5,12], coronary dilatation (2 cases) [5,9], coronary aneurisms (2 cases) [6,8] and perivascular brightening associated to other coronary wall abnormalities (1 case) [4]. Both cases of coronary aneurism occurred in children with delayed KD diagnosis after 10 or more days since fever onset.

4. Discussion

Kawasaki disease is an acute febrile illness characterized by a systemic vasculitis of small and medium sized arteries, that affects