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CASE REPORT OPEN ACCESS

TEMPORAL ABSCESS COMPLICATING OTOMASTOIDITIS: ANOTHER PRESENTATION OF KAWASAKI DISEASE

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ABSTRACT

Incomplete and atypical forms of Kawasaki disease increasingly pose a diagnostic challenge. We report a case of atypical presentation of Kawasaki disease with otomastoiditis complicated by temporal abscess and review the literature of this topic. A 4-year-old boy, presenting with Kawasaki disease, was treated with intravenous immunoglobulin and discharged after a week-long hospitalization. He presented with recurrence of fever and an acute evolving left temporal swelling. Computed Tomography scan showed left otomastoiditis with collection and infiltration of the temporal area. The oto-rhino-laryngological manifestations of Kawasaki disease are an integral component of the diagnosis. We found, in addition, a temporal abscess complicating otomastoiditis. All children with Kawasaki disease should have an oto-rhino-laryngological examination, especially if fever persists or recurs after treatment with immunoglobulin.

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INTRODUCTION

Kawasaki disease is the most common systemic vasculitis in children. 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 (Cavicchiolo *et al.,* 2012; Cimaz, 2007). Incomplete and atypical forms increasingly pose a diagnostic challenge. We report a case of Kawasaki disease with otomastoiditis complicated by temporal abscess and we analyze the oto-rhino-laryngological manifestations of Kawasaki disease in the literature.

Case report

A 4-year-old boy with no previous conditions, hospitalized a week ago for Kawasaki disease with prolonged fever (8 days), cheilitis, non-purulent bilateral conjunctivitis, cervical lymphadenopathy, ankle edema, maculopapular rash of the lower limbs and desquamation of the perineal region. The oto-rhino-laryngological examination was normal including no otitis or loco-regional swelling. The lab results showed an inflammatory syndrome. The echocardiography revealed an aneurism of the left coronary artery measuring 5mm. The patient was treated with intravenous immunoglobulin at the 5th day of hospitalization by the unavailability of immunoglobulin in our hospital (2g/kg) and acetylsalicylic acid on the 1st day of hospitalization (80mg/kg/day at first then 5mg/kg/day after immunoglobulin). The outcome was marked by the disappearance of the cutaneous signs and the fever after treatment, and the child was declared outgoing the next day of apyrexia. Two days after leaving the hospital, he presented with recurrence of the fever and appearance of an acute evolving left temporal pretragal swelling. The clinical examination noted a febrile child with a temperature at 38.9 °C, presenting a left temporal pretragal swelling of 7cm/5cm, tender, painful, with poorly defined contours, without trismus or signs of inflammation, with bilateral cervical lymphadenopathy measuring 3cm/2cm on the left and 2cm/2cm on the right. Ultrasonography showed significant infiltration of the left temporal region associated with a sub-periosteal collection of 23 mm and cervical lymphadenopathy. Left otomastoiditis with juxta-cortical collection measuring

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12mm/4mm and significant infiltration of soft tissue of the temporal area with lateral cervical lymphadenopathy was observed on cerebro-facial Computed Tomography scan (Figures 1 and 2). An inflammatory syndrome was found on the lab results, the C-Reactive Protein was 175 mg/l, the Erythrocyte Sedimentation Rate was 135 mm/h in the first hour, and leukocytosis at 26 600/mm3 predominantly neutrophilic at 21 120/mm3 with thrombocytosis at 629 000/mm3. The diagnosis of atypical Kawasaki disease was retained with the presence of a temporal abscess complicating otomastoiditis. The child was placed on antibiotics, amoxicillin-clavulanic acid for 10 days with spectacular clinical improvement and regression of swelling after 48 hours of treatment.

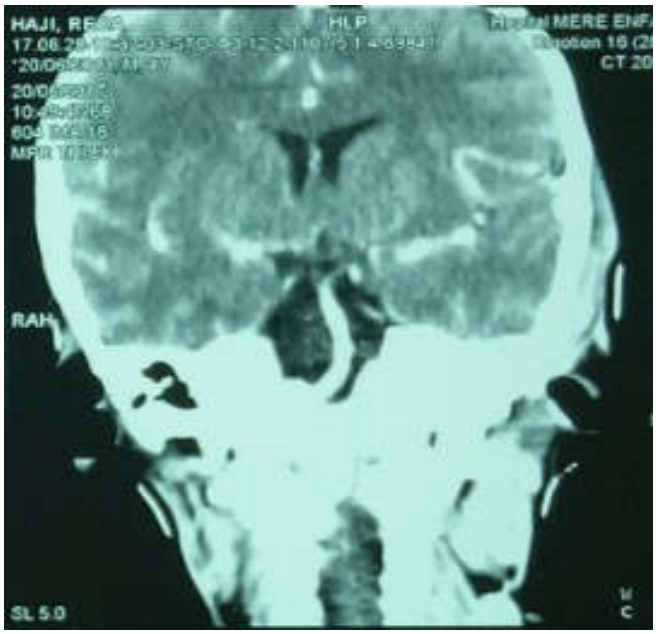

Figure 1.

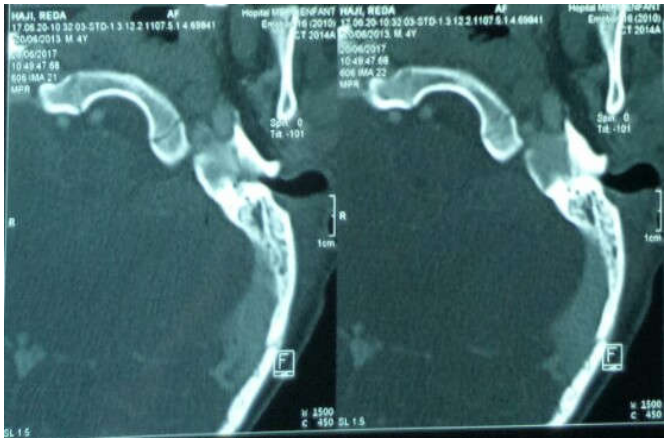

Figure 2.

DISCUSSION

Kawasaki disease is a febrile systemic vasculitis that, in the absence of treatment, is complicated by coronary aneurysms in 25 to 30% of cases. It is the most common cause of acquired heart disease in children in industrialized countries (Cimaz, 2007). It mainly affects children under 5, with a peak incidence in children aged 1 to 2 years. Rapid diagnosis is essential and treatment should be instituted as soon as the diagnosis of classical or incomplete Kawasaki disease is made (Cavicchiolo *et al.,* 2012 ; Cimaz, 2009), because the early administration of intravenous immunoglobulin associated with acetylsalicylic acid decreases the frequency of coronary artery abnormalities to less than 5%. The clinical picture comprises persistent fever, mucocutaneous signs (conjunctivitis, pharyngitis, strawberry tongue, cheilitis, polymorphous rash, desquamation of the extremities) and cervical lymphadenopathy. The pathogenesis of Kawasaki disease is still unknown, and several theories have been proposed, including the possibility of infection by toxin-secreting microorganisms and a process related to superantigens. Despite extensive research, there is still no diagnostic test available, and its diagnosis is based on clinical criteria after excluding other diseases with high and persistent fever (Cimaz, 2007; Watanabe *et al.,* 2014). The so-called "atypical" forms of Kawasaki disease should include patients with clinical manifestations that have been reported rarely in the syndrome (Cimaz, 2009). The symptoms of atypical Kawasaki disease are heterogeneous and may be associated; oto-rhino-laryngological diseases, neurological, severe bacterial infections, renal failure, pulmonary infiltrates, abdominal pain, arthritis and lymphadenitis. Due to the high frequency of head and neck manifestations, otolaryngologists play a central role in establishing the correct diagnosis of the disease. They look for peritonsillar abscesses, retropharyngeal abscess and acute otitis media (Cavicchiolo *et al.,* 2012; Kim, 2016). Head and neck manifestations of Kawasaki disease, including conjunctivitis, changes in oral mucosa and, less frequently, cervical lymphadenopathy, are integral components of the diagnosis. In addition, there are many other symptoms of the disease including common entities such as deep neck abscess, aseptic meningitis, torticollis, facial nerve palsy and rhinitis. Other symptoms such as airway obstruction, infected branchial cleft cyst, necrotizing pharyngitis and sensorineural hearing loss are rare (Yoskovitch, 2000). Moreover, we found a temporal abscess complicating otomastoiditis, exceptionally described in the literature. In patients with Kawasaki disease, if the symptoms do not resolve even after intravenous immunoglobulin therapy, the coexistence of an oto-rhino-laryngology sphere infection including parapharyngeal or retropharyngeal abscess should be considered (Choi, 2010). Our patient; was treated with intravenous immunoglobulin before being readmitted for recurrence of fever and left temporal abscess secondary to otomastoiditis, without any history of inflammatory involvement of the oto-rhino-laryngological sphere.

Conclusion

All children with Kawasaki disease should be given oto-rhino-laryngological examination, especially in case of recurrence or persistence of fever after treatment with intravenous immunoglobulin. The oto-rhino-laryngologists should exclude otomastoiditis or temporal abscess even in the absence of a specific signs.

Conflict of interest: None

Consent for publication: A written informed consent was obtained from all patients when they were enrolled.

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