Kawasaki Disease presenting with intussusception: a case report

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Abstract
A 3 yr old boy presented with abdominal pain, fever and red jelly stools. Intussusception was diagnosed and effectively reduced with air insufflation. However, despite an improvement in his clinical condition, the child remained febrile and miserable; 5 days later he developed characteristic signs of Kawasaki disease and was treated with intravenous immunoglobulin and high dose aspirin with good results. Intussusception prior to the typical features of Kawasaki disease has not been described previously in the English literature. This case illustrates a novel presentation of Kawasaki disease.

Case
A 3 yr old Caucasian boy was referred from the accident and emergency department to the paediatric surgical team with a history of acute onset abdominal pain. The pain was central, and associated with fever, non bilious vomiting and the passage of red, jelly-like stools. There was no history of weight loss, jaundice or foreign travel; there was no contact history and no recent change in dietary habits. Other than a mild upper respiratory tract infection, the child was previously fit and well with no relevant medical history. On examination, the child was well nourished; he was miserable and lethargic with fever (38.1°C) and tachycardia (heart rate 120/min). He was warm and well perfused with a capillary refill time of <2 seconds both centrally and peripherally. His abdomen was tender, distended and resonant; bowel sounds were hyperactive and a small, tender, rubbery mass was palpable centrally. An ultrasound scan was undertaken and confirmed the presence of an intussusception. The child underwent a contrast enema; control films showed a filling defect in the ascending colon at the location of the colo-colic intussusception. During a subsequent air enema, air insufflation to a pressure of 120 mmHg was sufficient to reduce the intussusception adequately. The child was kept in hospital overnight for observation.

By the following day, the boy’s condition had improved with resolution of his abdominal pain; he was able to eat and drink and interact with his parents and staff. However he continued to have a remittent fever to 38.5°C and he remained lethargic and miserable. Subsequently, he developed a diffuse morbilliform rash over the trunk and limbs and cervical lymphadenopathy was noted with an associated congested throat and enlarged tonsils, which was thought to be viral in origin. Blood tests revealed a C-reactive protein 61 mg/L, normal blood count, serum electrolytes, liver function tests and chest X-ray; Ebstein Barr viral serology, blood cultures and urine cultures were negative. Five days after initial presentation, he developed conjunctival oedema and oedema of the hands and feet with desquamation of the finger tips and cracking of the lips; there was no evidence of a strawberry tongue. Kawasaki Disease was diagnosed. The child was treated with a dose of intravenous immunoglobulins (2 g/kg) and initiated on a therapeutic high dose aspirin (25 mg/kg QDS). He made a good recovery and was discharged 4 days later. The aspirin was altered to a lower dose (5 mg/kg OD) after 2 weeks following an initial normal echocardiogram. Echocardiography was repeated at 8 weeks and showed no coronary artery abnormality. The aspirin was subsequently stopped.

Discussion
Intussusception is the commonest cause of intestinal obstruction in infants, and occurs with an incidence of approximately 66/100,000 in the UK [1]. Lead points may include polyps, lymph nodes, diverticulae. However in most cases the cause is unknown and is thought to be due to enlarged Peyers patches; enlarged intestinal
lymph nodes have been described in those requiring surgical intervention[2,3]. Intestinal lymphadenopathy can occur for many reasons such as Crohn's disease, tumours, gastrointestinal infections or drug reactions; however enlarged Peyer's patches occur most often in young children as a reaction following viral upper respiratory tract or gastrointestinal infection. Intussusception is therefore unsurprisingly more common in the spring and autumn seasons[4]. This theory is further strengthened by reports of an increasing incidence of intussusception following the introduction of the rotavirus vaccination in the US[5].

Kawasaki Disease is similarly postulated to be triggered by an initial infective insult, with a similar preceding history of a viral upper respiratory or gastrointestinal infection, and similar seasonal pattern of disease[6]. On presentation, a proportion of patients (approximately 40%) exhibit cervical lymphadenopathy; however whether children have concurrent lymphadenopathy elsewhere is unknown. Development of intestinal mucosal lymphadenopathy due to the same trigger viral pathogen may explain the process by which intussusception occurs in these children.

Gastrointestinal symptoms are common in Kawasaki disease; abdominal pain, nausea and diarrhoea are seen in up to one third of cases, and these are therefore often regarded as symptoms of Kawasaki disease rather than of potential intra abdominal complications. Symptoms often resolve on treatment with intravenous immunoglobulins. This child also developed coronary artery aneurysms. There has been just one case, a 3 month old boy[10], initially presenting with symptoms of Kawasaki disease and was thereby subsequently treated with immunoglobulins. This child also developed coronary artery aneurysms.

This is the first report in the English literature of a child presenting with an intussusception with no initial symptoms of Kawasaki disease. The two conditions were highly likely to have been associated in this case. It is important to consider underlying causes of intussusception in this older age group of children, and consider Kawasaki disease in those febrile on presentation. We wish to highlight this novel presentation of Kawasaki disease.

Consent
Written informed consent was obtained from the patient's parents for publication of this case report. A copy of the written consent is available for review by the Editor-in-Chief of this journal.

Abbreviations

CRP: C-reactive protein

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Authors' contributions

All authors read and approved the final manuscript.

Competing interests

The authors declare that they have no competing interests.

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