Adult Burkitt's lymphoma presenting as intussusception: first UK case report

R Simson, A Planner, Roderick Alexander

Great Western Hospital, Swindon, Wiltshire, UK

ABSTRACT

Colonic intussusception is rare in adults and can present with non-specific symptoms that can make diagnosis difficult and delayed. Unlike in children, it is commonly due to a pathological lead point that is often malignant. This case is the first reported case of adult intussusception due to Burkitt's lymphoma in the UK. We describe the case of a 22-year-old woman who presented with 4-week history of intermittent epigastric pain. On the third hospital admission, the diagnosis was made by abdominal computed tomography, which showed the 'target' sign, suggestive of intussusception. A right hemicolectomy was performed and histology later confirmed Burkitt's lymphoma. This case demonstrates the difficulty in diagnosing intussusception in adults, which must be considered in recurrent abdominal pain when more common causes have been ruled out. The rare diagnosis of Burkitt's lymphoma made early diagnosis and treatment important.

KEYWORDS

Intussusception – Burkitt's lymphoma – Abdominal pain – Adult – Early diagnosis

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CORRESPONDENCE TO

Rosie Simson, E: rosiesimson@hotmail.co.uk

Introduction

This is the first reported case of Burkitt's lymphoma presenting as intussusception in the UK in an adult: indeed there are only 15 reported cases worldwide.^{1,2} Only 5% of all cases of intussusception occur in adults, in whom it accounts for 1% of cases of intestinal obstruction.³ Unlike in children, intussusception in adults has a pathological lead point in 70–90% of cases. The lead point is malignant in up to 65% of cases.^{3,4}

Case Report

A 22-year-old woman presented with 4-week history of intermittent epigastric pain, unrelated to eating with no other gastrointestinal or systemic symptoms. On examination, there was mild epigastric tenderness and marked tenderness in the right iliac fossa with guarding. Her C-reactive protein (CRP) level was 93mg/l (range 0–6mg/l), white cell count 10.4 (range 4–10 × 10⁹/l) and alanine transaminase 58 u/l (range 5–45 u/l). She underwent a diagnostic laparoscopy and appendicectomy performed by a year 7 specialist trainee registrar. A small amount of clear fluid was noted in the pelvis. The appendix had congenital adhesions but was normal in appearance. Subsequent histology confirmed a macroscopically and histologically normal appendix. There

was no evidence of intussusception at the time of laparoscopy.

The patient returned 4 days postoperatively with abdominal pain, vomiting and loose stools. On examination, she was tender in the central abdomen and right iliac fossa. Bloods results showed raised a CRP level of 15mg/l but were otherwise normal. An ultrasound of the abdomen was performed by an experienced radiological registrar, which did not show any collections of fluid and was reassuring. oesophago-gastroduodenoscopy showed gastritis and, as her symptoms had settled, she was discharged with a proton pump inhibitor.

She was readmitted 2 weeks later with recurrent and worsening epigastric pain associated with nausea and vomiting. Blood results showed a white cell count of 10.5 × 10⁹/l and raised CRP of 23mg/l. Because of her continuing symptoms, a plain abdominal x-ray was taken, which showed no significant findings or obstructive pattern but, with hindsight, a subtle central opacity is observed (Fig 1). Computed tomography (CT) of her abdomen and pelvis showed colonic intussusception with enlarged mesenteric lymph nodes (Figs 2 and 3). Laparotomy confirmed ileocolic intussusception and a right hemicolectomy was performed. Histology showed a 5.5cm necrotic mass in the terminal ileum as a lead point of the intussusception. Microscopy showed atypical cells with a starry sky background and foamy histiocytes.

Figure 1 Plain abdominal x-ray performed prior to computed tomography showing no definite abnormality or small bowel obstruction.

Immunohistochemistry confirmed Burkitt's lymphoma. Subsequent HIV tests were negative and she was treated with chemotherapy.

Discussion

Burkitt's lymphoma is a rapidly growing B-cell neoplasm. Its estimated incidence in patients 20–30 years of age is 0.6 per million.⁵ In endemic areas, Burkitt's lymphoma usually affects the maxilla and presents with swelling of the affected bones.² In the sporadic form, where distal ileum, caecum and mesentery are most frequently affected, presenting symptoms include abdominal swelling due to large mesenteric retroperitoneal or pelvic mass, abdominal pain and intussusception-related symptoms of bowel obstruction.² Intussusception in itself is a very rare presentation of Burkitt's lymphoma. In this case there was no abdominal mass felt; however, it is well documented in other case studies.¹

Intussusception is uncommon in adults.⁴ The clinical triad of vomiting, rectal bleeding and abdominal pain seen in children with intussusception is only present in 15–20% of adults,⁵ in whom the presentation is typically with non-specific symptoms including abdominal pain and nausea. The absence of acute obstruction can delay the diagnosis, as in our case.³ Awareness of the possibility of intussusception

Figure 2 Coronal abdominal computed tomography: lead point mass at level of ileocaecal valve (blue arrow); adjacent lymph nodes within fatty mesentery (yellow arrow); small bowel being held up at point of intussusception (green arrow); concertina effect of right colon bowel wall within transverse colon (red arrow).

can lead to earlier diagnosis with CT.³ In adults, CT can aid both diagnosis and assessment of pathological lead points and thus is the imaging of choice in adult intussusception.⁴

It is most commonly secondary to a lead point of variable pathology including gastrointestinal stromal tumour, lipoma, Meckel's diverticulum and metastatic melanoma.⁵ In the small bowel, 20% of intussusceptions are idiopathic.⁴ Intussusception in the large bowel is more likely to have a malignant aetiology and represents up to two-thirds of cases, with adenocarcinoma and lymphoma being the most common.⁴

Conclusion

It is probable that the recurrent abdominal pain and vomiting in this patient was due to the intussusception that was not detected during the first two admissions. We speculate that, given the absence of intussusception at the time of laparoscopy and resolution in symptoms seen, the intussusception, to begin with, was intermittent.

This case demonstrates the difficulty in diagnosing intussusception in adults, which must be considered in recurrent abdominal pain when more common causes have been ruled out. The high proportion of malignant lead points, as in our case, makes early diagnosis and treatment important.

Figure 3 Abdominal computed tomography: sagittal images demonstrating typical 'target sign' of intussusception in proximal transverse colon.

Furthermore, the use of CT in the context of the clinical setting is helpful in diagnosing intussusception and identifying the lead point early.

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