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CASE REPORT

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Neutropenic enterocolitis (typhlitis) associated with infectious mononucleosis

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Abstract Neutropenic enterocolitis (typhlitis) is an unusual acute complication of neutropenia, most often associated with leukaemia and lymphoma and characterized by segmental caecal and ascending colonic ulceration that may progress to necrosis, perforation, and septicaemia. We present a unique case of an 8-year-old girl with recently diagnosed infectious mononucleosis having findings consistent with typhlitis on abdominal CT.

Keywords Colon · Caecum · Infection · Typhlitis · Child · CT

Introduction

Typhlitis, also known as neutropenic enterocolitis, is an unusual acute complication of neutropenia [1, 2]. We present an 8-year-old girl with recently diagnosed infectious mononucleosis complicated by typhlitis. To the best of our knowledge, neutropenic enterocolitis as a complication has not previously been described in infectious mononucleosis.

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Case report

An 8-year-old girl presented to another hospital with abdominal pain, diarrhoea, weight-loss and pallor. Trimethoprim-sulfamethoxazole, metronidazole and antiinflammatory drugs were prescribed. She came to our hospital after failing to improve and having developed additional fever and neck swelling. She had also decreased hearing.

Laboratory evaluation revealed pancytopenia: haemoglobin 6.3 g/dl, haematocrit 18.4%, leucocytes 400/mm^3, platelets 97,000/mm^3. Bone marrow aspiration showed reduced premyeloid cells. *Escherichia coli* was grown from blood culture and this prompted the addition of ceftazidime to the treatment regimen; filgrastim was added because of the neutropenia. The fever continued and so vancomycin and meropenem were added. Sonography and CT (Fig. 1) of the neck revealed multiple lymph nodes, the largest measuring 2×1.6 cm. Cervical lymph node aspiration revealed positive Epstein-Barr virus (EBV) with EBV-VCA IgG(+), EBV-VCA IgM(−), EBV PCR(+), CMV PCR(−). Abdominal US showed mild splenomegaly, increased caecal and proximal ascending colon wall thickness. Abdominal CT (Fig. 2) revealed diffuse thickening of the caecal and ascending colonic walls with air-fluid levels and linear inflammatory stranding of the pericaecal fat. The diagnosis of typhlitis was made. Acyclovir was prescribed together with metronidazole because the patient came from an area that is endemic for amoebiasis and also had ongoing diarrhoea. After 10 days, repeat CT demonstrated that the imaging findings compatible with typhlitis had significantly regressed.

Discussion

Typhlitis, also known as neutropenic enterocolitis or ileocaecal syndrome, is an unusual acute complication of neutropenia, most often associated with leukaemia and lymphoma and characterized by segmental caecal and as-

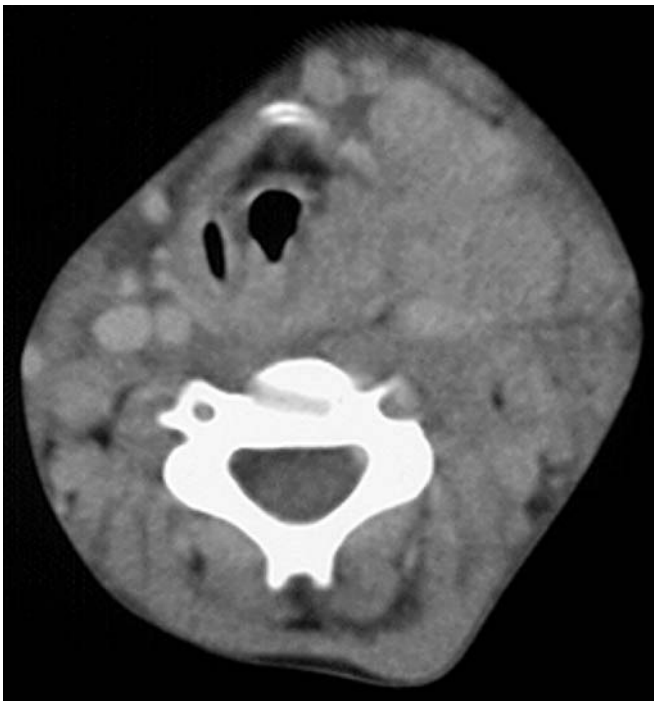

Fig. 1 Enhanced neck CT shows lymphadenopathy especially in the left side of the neck

cending colonic ulceration that may progress to necrosis, perforation and septicaemia. However, typhlitis has also been reported in patients with aplastic anaemia and acquired immunodeficiency syndrome, and after renal transplantation [1, 2]. Infectious mononucleosis, or glandular fever, is a condition caused by EBV but as many as 5–10% of infectious mononucleosis-like illnesses are caused by cytomegalovirus, adenovirus, *Toxoplasma gondii*, HIV, and possibly rubella virus [3]. To our knowledge, there has been no case report in the literature of typhlitis associated with infectious mononucleosis in the paediatric age group.

Typhlitis is characterized by oedema and inflammation of the caecum, the ascending colon and sometimes the terminal ileum. Inflammation can be so severe that transmural necrosis, perforation and death can result. The mechanism of the condition is not known, but is probably due to a combination of ischaemia, infection (especially with cytomegalovirus), mucosal haemorrhage, and perhaps neoplastic infiltration [2]. Patients present with fever, diarrhoea and abdominal pain that may be localized to the right lower quadrant. Infectious mononucleosis usually causes mild thrombocytopenia (50,000–200,000 platelets/mm^3) and neutropenia, but severe thrombocytopenia (<20,000 platelets/mm^3) and severe neutropenia (<1,000 neutrophils/mm^3) are rare [3]. In our patient, mild thrombocytopenia (97,000 platelets/mm^3) and severe neutropenia (400 neutrophils/mm^3) were present. Neutropenia, and hence the lack of normal granulocyte reaction to infection, relative stasis of bowel contents in the caecum, and the distensibility of the caecum with compromise of its blood supply, may predispose the caecum to the site of typhlitis [4].

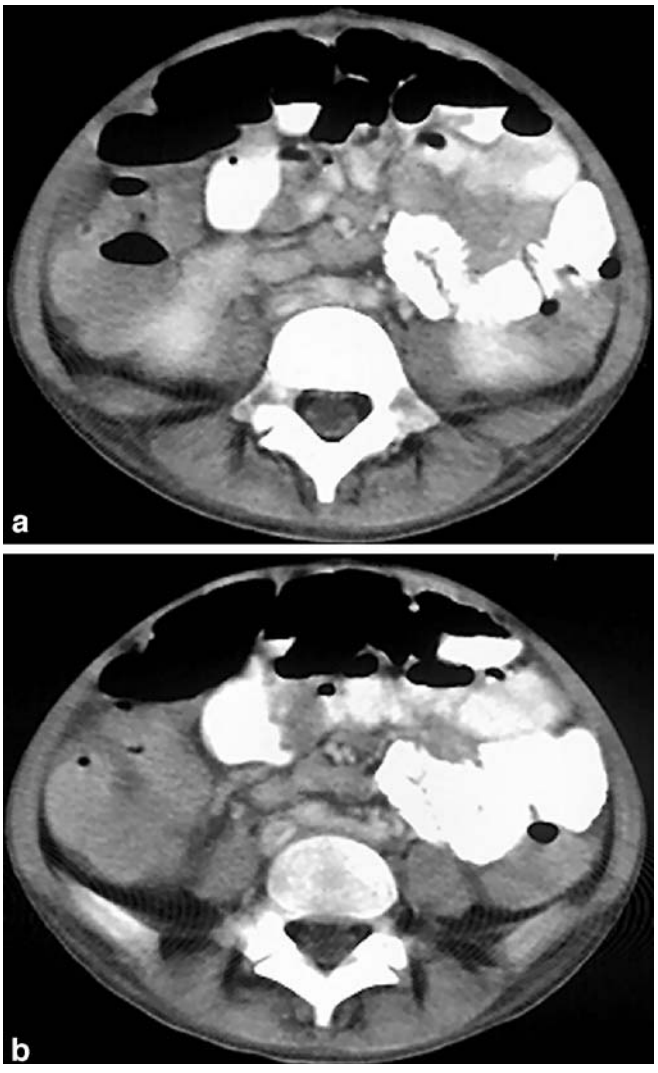

Fig. 2 Contrast-enhanced abdominal CT shows thickening of the caecal wall with pericaecal inflammatory stranding (**a**)

Diagnosing typhlitis can be difficult. CT is the study of choice for the diagnosis of typhlitis due to the absent risk of bowel perforation when compared with colonoscopy or contrast enema examination. CT reveals a thickened caecal wall and caecal distension [5] (Fig. 2). Inflammatory stranding of the adjacent mesenteric fat is a common finding (Fig. 2). Sonography is also an excellent imaging modality, particularly if the patient is too ill to be moved to the scanner [6]. Suarez et al. [7] and Alexander et al. [8] have reported the US findings in four cases of typhlitis and concluded that US offers a noninvasive method for diagnosing and monitoring this potentially lethal disease. However, US depends on the experience of the sonographer and is also affected by a significant ring-down artefact that is caused by distended right-lower quadrant small-bowel loops obscuring visualization of the right colon. Sonography may also be inadequate to diagnose the main reasons for mortality—caecal perforation, bowel necrosis, and abscess formation. CT readily and effectively identifies

these complications as well as pneumatosis intestinalis, pneumoperitoneum, ascites and abscess that indicate the need for urgent surgical intervention [5, 9]. CT may also be indicated to assess the response to treatment (as in our patient) and to establish a baseline to compare follow-up studies [2].

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