Simultaneous double ileoileal intussusception due to Burkitt’s lymphoma in a young male

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Burkitt’s lymphoma is an uncommon cause of intussusception in adults. Double intussusceptions due to Burkitt’s lymphoma are extremely rare. We present a case of a 26-year-old man who presented with symptoms of intestinal obstruction and was diagnosed with double ileoileal intussusception at laparotomy. The pathology of the lead points turned out to be Burkitt’s lymphoma. This could be the first reported case in the literature.

INTRODUCTION

Intussusception is the invagination of a bowel loop with its mesenteric fold (intussusceptum) into the lumen of a contiguous portion of bowel (intussuscipien) as a result of peristalsis [4]. Intussusception is the invagination of a bowel loop with its mesenteric fold (intussusceptum) into the lumen of a contiguous portion of bowel (intussuscipien) as a result of peristalsis [4].

CASE REPORT

A 26-year-old male patient, previously healthy, presented to our ER complaining of diffuse colicky abdominal pain of one week duration. He also reported nausea, two episodes of non-bilious vomiting and a two-day history of constipation and obstipation. No fever or chills. He denied any past surgical history. His vital signs were all within normal limits. On physical examination, his abdomen was tender, distended and tympanic on percussion. Bowel sounds were hyperactive. DRE showed empty rectum. All his lab tests were normal except for mild leukocytosis. Plain abdominal film showed two air-fluid levels. A follow-up CT scan of the abdomen and pelvis showed small bowel obstruction and a 5 × 5 cm mesenteric mass (Fig. 1). Another mass of 3 × 3 cm was also noted. The patient was referred for surgical management.

DISCUSSION

Burkitt’s lymphoma is an uncommon cause of intussusception in adults. Double intussusceptions due to Burkitt’s lymphoma are extremely rare. We present a case of a 26-year-old man who presented with symptoms of intestinal obstruction and was diagnosed with double ileoileal intussusception at laparotomy. The pathology of the lead points turned out to be Burkitt’s lymphoma. This could be the first reported case in the literature.

Under general anesthesia, patient in supine position, a midline laparotomy was performed. A large amount of abdominal fluid was aspirated and sent for culture and cytology. During running of the bowel at the level of 20 cm from the ligament of Treitz, an intussusception with a palpable intraluminal mass was found. Another intussusception 120 cm distal to the first was also noted (Fig. 2). No other pathologies were found. Double enterectomies with end-to-end anastomoses were performed. The patient was discharged home on Day 5 post op. Pathological analysis revealed polypoid and infiltrating intraluminal lymphoid proliferation (Fig. 3), extending to serosa and exhibiting diffuse aspect composed of medium-sized B-cells: pattern of Burkitt’s Lymphoma. Abdominal fluid cytology analysis showed the presence of atypical lymphoid cells.
hospital admissions [4] and 1/1300 of all abdominal operations [5]. Unlike in infancy, 70–90% of adult intussusceptions are due to some type of intestinal lesion [4]. Table 1 presents a comparison between pediatric and adult-type intussusceptions.

Box 1 outlines some of the rare reported causes of intussusceptions [5].

| Ca appendix | Adenomyoma in a Meckle’s diverticulum |
| Submucous lipoma | Extramedullary haematopoietic tumor |
| Haemangioma of small bowel | Endometriosis of terminal ileum |
| Peutz Jegher’s polyp | Metastatic testicular germ cell tumor |
| ‘Vanished’ colonic tumor with deposits in glands | Pneumatosis coli |
| Coeliac disease | Ileal aberrant pancreas |
| Duodenal villous adenoma | Metastatic melanoma of ileum |
| Bowel wall haematoma | Gastroduodenal due to gastric carcinoma |
| Others |

There are several proposed tools to aid in the diagnosis of intussusceptions. However, CT scan remains the most useful radiologic method for diagnosing intestinal intussusception according to several studies [2, 4]. Treatment of adult intussusceptions is usually surgical. Some advocate reduction of the intussusceptions, whereas others prefer resection without reduction, hence decreasing the likelihood of bowel injury or ischemia, and the dissemination or perforation of malignancy.
However, Horton KM et al. found that intussusception is increasingly being detected by MDCT, even in asymptomatic patients, and given the recognition that intussusceptions may be transient, there is ongoing controversy regarding the optimal management, and have recommended that in asymptomatic patients, the CT identification of a proximal (jejunal), short (<3.5 cm) intussusception with a characteristic target sign and without obstructive symptoms may indicate a self-limiting process that can be managed conservatively. [6]. Double intussusception is extremely rare. There are four subtypes [3]:

(i) Two separate intestines prolapsing into the same distal intestine, resulting in a characteristic ‘triple-circle’ sign on abdominal sonography and CT scan.
(ii) The double compound intussusception, which is extremely rare being reported once in the literature.
(iii) The double prolapse of the proximal and distal intestine through a patent vitello-intestinal duct.
(iv) Double-site intussusceptions, like the case we have presented.

Previous reports have shown that up to 50% of adult intussusceptions are due to intestinal neoplasms, including malignant lymphoma [4, 5].

The gastrointestinal tract is the most common extranodal site of lymphoma [7]; Pathologically, Burkitt’s lymphoma and enteropathy-associated T-cell lymphoma, with MALT type are the most common lymphoma of the small intestine and account for 42.5% of the lymphomas [8]. Burkitt’s lymphoma is a diffuse, undifferentiated, malignant monoclonal B-cell lymphoma with two major clinical presentations. American Burkitt’s differs from the African type described by Burkitt in 1958 in its increased propensity for widespread involvement, especially within the abdominal cavity [9].

Surgery, chemotherapy, radiotherapy and radioimmunotherapy are the different modalities for the management of GI lymphoma and can be applied in different combinations [7]. Complete tumor resection with minimal complication is possible in most patients who present with intussusception, thus downgrading their stage of disease, hence requiring shorter duration and less intense chemotherapy minimizing the risk of early and late complications. [10].

According to the data from US SEER data for the period 1992–2005, the 5-year relative survival was 64.1% for lymphomas [8].

Hereby we presented to you a case of adult double intussusception due to Burkitt’s lymphoma. There are many cases of intestinal lymphomas presenting with intussusception. To our knowledge, this is the first case of Burkitt’s lymphoma presenting with intestinal obstruction due to simultaneous double intussusception to be reported.

REFERENCES

1. Eisen LK, Cunningham JD, Aufses AH, Jr. Intussusception in adults: institutional review. J Am Coll Surg 1999;188:390–5.
2. Krasniqi AS, Hamza AR, Salihu LM, Spahija GS, Bicaj BX, Krasniqi SA, et al. Compound double ileoileal and ileocecocolic intussusception caused by lipoma of the ileum in an adult patient: a case report. J Med Case Rep 2011;5:452.
3. Chen Y, Diau G, Chang C, Chen K, Chu C. Double site intussusception in a four-year-old girl. J Med Sci 2006;26:191–4.
4. Azar T, Berger DL. Adult intussusception. Ann Surg 1997;226:134–8.
5. Rathore MA, Andrabi SIH, Mansha M. Adult intussusception—a surgical dilemma. J Ayub Med Coll Abbottabad 2006;18(3).
6. Horton KM, Fishman EK. MDCT and 3D imaging in transient enteroenteric intussusception: clinical observations and review of the literature. AJR Am J Roentgenol 2008;191:736–42.
7. Prasanna Ghimire, Guang-Yao Wu, Ling Zhu. Primary gastrointestinal lymphoma. World J Gastrointest Oncol 2011;3:33–42.
8. Pan SY, Morrison H. Epidemiology of cancer of the small intestine. World J Gastrointest Oncol 2011;3:33–42.
9. Carbone P, Berard CW, Bennett JM, Ziegler JL, Cohen MH, Gerber P. NIH Clinical Staff Conference—Burkitt’s tumor. Ann Intern Med 1969;70:817.
10. Gupta H, Davidoff AM, Pui CH, Shochat SJ, Sandlund JT. Clinical implications and surgical management of intussusception in pediatric patients with Burkitt lymphoma. J Pediatr Surg 2007;42:998–100.