CASE REPORT

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Adult small bowel intussusception secondary to Burkitt lymphoma

Bertrand Ng Ren Joon, Yasser Arafat, Ishtiaq Alam, Vishvendra Gaur

ABSTRACT

Introduction: Intussusception results from telescoping of proximal gastrointestinal segment into a distal segment. It is infrequent cause of bowel obstruction in adults in comparison to children. Hence, it is important for surgeons to be aware of this condition in adults presenting with symptoms of bowel obstruction. We present a rare case of an adult small bowel intussusception secondary to Burkitt lymphoma along with a literature review. Case Report: A 69-year-old gentleman presented to the emergency department (A&E) with two days history of generalized colicky abdominal pain along with symptoms of obstipation. There is no previous history of abdominal surgery. A computed tomography (CT) abdomen and pelvis demonstrated an enteroenteric intussusception in the right mid abdomen resulting in small bowel obstruction. He underwent an emergency laparotomy. Intraoperatively, there was an enteroenteric intussusception and he had a resection of the segment. Histology revealed a non-Hodgkin high grade B cell lymphoma. He was well after and was transferred to hematology team for further treatment. **Conclusion:** Intussusception in adult is rare and could prove challenging for clinician as some diagnoses are only made intraoperatively. Awareness and early recognition of Burkitt lymphoma as a possible differential diagnosis in adult with intussusception is essential as further

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Keywords: Adult, Burkitt lymphoma, Enteroenteric, Intussusception, Laparotomy

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INTRODUCTION

Intussusception can affect patient at any age, but it is more common in children between 5 and 10 months of age [1]. Its occurrence in adult is rare with approximately 2–3 cases per 1 million population per year and occurrence in adult has an equal male to female prevalence [1, 2]. Diagnosis of intussusception sometimes is only made intraoperatively and may prove to be a challenge for the surgeon. Burkitt lymphoma, on the other hand, is an aggressive, rapidly dividing B cell cancer that occurs rarely in adult too. Timely diagnosis and treatment in Burkitt lymphoma patient are essential as it could result in improved survival rate. We present a rare case of adult small bowel intussusception who underwent a small bowel resection with primary anastomosis and a confirmatory diagnosis of Burkitt lymphoma on histology was made.

CASE REPORT

A 69-year-old gentleman presented to the A&E with two days history of acute abdominal pain followed by



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multiple episodes of vomiting and fever. On arrival to A&E he was hemodynamically unstable with hypotension and tachycardia.

He did not have any significant background medical history and any previous surgical history.

His abdominal examination revealed localized sign of peritonism. Apart from raised serum lactate 2.45 mmol/L and deranged LFT with ALT 174 U/L and AST 221 U/L, the rest of the blood results were within normal limit. Computed tomography scan of abdomen and pelvis demonstrated an enteroenteric intussusception in the right mid abdomen resulting in partial bowel obstruction with mild dilated loops of proximal small bowel (Refer Figures 1 and 2). There were no evidence of perforation. No mass lesion was clearly identified within the bowel; however, an incidentaloma in the right adrenal gland was found, measuring 48 × 62 mm. The liver, gallbladder, pancreas, spleen, and both kidneys were unremarkable. There was no periaortic or mesenteric lymphadenopathy noted.

He underwent an emergency laparotomy and intraoperatively there was an irreversible invaginated loop of small bowel, for which he underwent resection of the enteroenteric intussusception at 120 cm proximal to ileocecal junction (Figures 3–5). A side-to-side staple anastomosis was performed. The peritoneal survey, rest of small bowel and liver were unremarkable.

The histology demonstrated a 35 cm poorly differentiated non-Hodgkin high grade B cell lymphoma which was positive for CD 20, BCL 6, c-MYC with a high proliferative index (close to 100%) and rearranged c-MYC on fluorescence in situ hybridization (FISH) evaluation. Microscopically, there were predominantly intermediatesized lymphocytes with vesicular chromatin and small nucleoli and in some area, blast-like lymphocytes were present. BCL-2 and CD-10 were negative. Although the

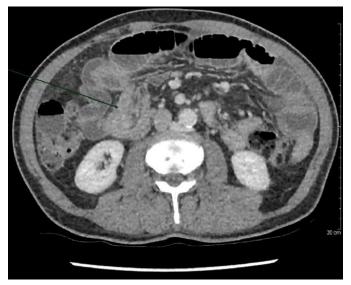


Figure 1: Axial view of CT abdomen pelvis. Arrow showing enteroenteric intussusception in the right mid abdomen with mildly dilated loops of proximal small bowel.



Figure 2: Sagittal view of CT abdomen pelvis of the enteroenteric intussusception.

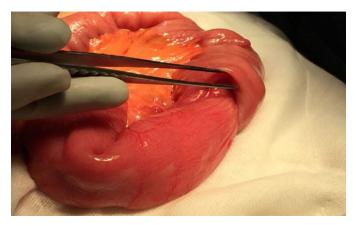


Figure 3: Close up view of the enteroenteric intussusception.

CD-10 was absence, the clinical features, morphology, immunophenotype, and FISH results were typical of sporadic Burkitt lymphoma, hence, a diagnosis of Burkitt lymphoma was made and he was transferred to the hematology unit for further management. He was commenced on allopurinol and prednisolone. After his discharge from the ward, he completed 6 cycles of R-CHOP chemotherapy and has been in remission since. He was referred to endocrine team for further follow-up of his incidentaloma.

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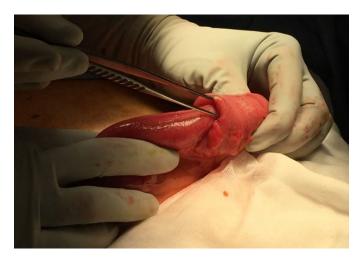


Figure 4: Intraoperative findings of the enteroenteric intussusception.

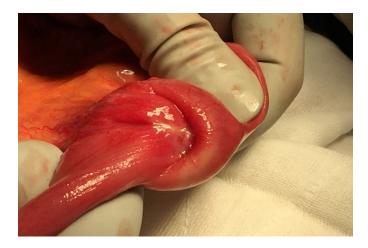


Figure 5: Intussusception prior to small bowel resection and anastomosis.

DISCUSSION

Intussusception occurs when a segment of the gut telescopes into an immediate adjacent segment. It occurs most commonly in children with peak incidence between 5 and 10 months [3]. Children classically present with intermittent colicky abdominal pain and vomiting with some occasionally presenting with abdominal distension, palpable sausage mass on palpation or passage of red currant jelly stool. On the other hand, bowel intussusception is rare in adults, comprising of 5% of cases of intussusception and only 1-5% of cases of mechanical small bowel obstruction [4]. One of the retrospective study review done by Zubaidi stated that abdominal pain, nausea, and vomiting were the commonest symptoms in adult intussusception, with 86% of adult cases associated with a definable lesion, and 29% of enteric lesion were found to be malignant [5]. Intussusception can be classified based on the anatomical location of the intussusception with ileocolic

being the most common in children and colocolic in adult [3].

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Burkitt lymphoma is a highly aggressive B cell non-Hodgkin lymphoma with three main clinical variants: endemic, sporadic, and immunodeficiency and they differ in terms of epidemiology, clinical presentation, and genetic features [6]. The exact worldwide incidences of Burkitt lymphoma are not known as there are limited data around. The endemic type is mainly found in Africa and New Guinea, with jaw/facial bone tumor noted in 50-60% of cases and nearly all are associated with Epstein-Barr virus infection [7, 8]. The sporadic variant involves 30% of pediatric lymphoma and <1% of non-Hodgkin lymphoma cases in the United States and they commonly present with abdominal symptoms which comprise of bowel obstruction or gastrointestinal bleeding [8, 9]. Immunodeficiency variant is commonly seen in human immunodeficiency virus (HIV)-infected immunodeficiency patient. Burkitt lymphoma alone in adult is rare with small bowel intussusception secondary to Burkitt lymphoma being even more extremely rare.

Over the years, CT scan has increasingly become a modality of investigation choice for acute abdomen patient. Computed tomography allows for visualization of both mural and extramural of small bowel, regional lymphadenopathy, and bowel wall mass and will help in differentiating lymphoma from other bowel disease [10]. Computed tomography scan has been the most useful imaging tool for adult intussusception with diagnostic yield around 78% and can also be helpful in identifying a lead point [11].

Invariably, the treatment for this disease is resection and primary anastomosis with involved bowel segment in adults who are diagnosed with pathological intussusception [12]. There is still controversy as to whether reduction of intussusception is required at operation with some reports suggesting that cancer cells might disseminate while attempting reduction, with the advantage remained to be to preserve a considerable length of bowels in order to prevent short gut syndrome [13]. In a study conducted by Demirkan et al., one of the major differences between adult and children intussusception is that adults are typically associated with underlying pathology and in their study no adult patient underwent nonoperative reduction, whereas pediatric patients were managed either with hydrostatic reduction or surgery [14].

CONCLUSION

Intussusception in adult is rare and could prove challenging for clinician as some diagnoses are only made intraoperatively. Awareness and early recognition of Burkitt lymphoma as a possible differential diagnosis in adult with intussusception is essential as further imaging and early referral to hematology unit could confer early intervention, such as chemotherapy to the patient.

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Drafting the work, Final approval of the version to be published, Agree to be accountable for all aspects of the work in ensuring that questions related to the accuracy or integrity of any part of the work are appropriately investigated and resolved

Yasser Arafat - Conception of the work, Analysis of data, Interpretation of data, Revising the work critically for important intellectual content, Final approval of the version to be published, Agree to be accountable for all aspects of the work in ensuring that questions related to the accuracy or integrity of any part of the work are appropriately investigated and resolved

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Vishvendra Gaur – Conception of the work, Acquisition of data, Analysis of data, Interpretation of data, Revising the work critically for important intellectual content, Final approval of the version to be published, Agree to be accountable for all aspects of the work in ensuring that questions related to the accuracy or integrity of any part of the work are appropriately investigated and resolved

Guarantor of Submission

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Written informed consent was obtained from the patient for publication of this article.

Conflict of Interest

Authors declare no conflict of interest.

Data Availability

All relevant data are within the paper and its Supporting Information files.

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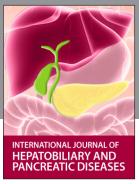
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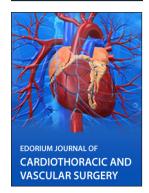














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