

Case Report

Intestinal Intussusception Secondary to Burkitt's Lymphoma in an Adult: A First Case Report in Latin America

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Abstract

Introduction: Intestinal intussusception is a pathology in which an intestinal segment and its mesentery are telescoped in the lumen of an adjacent intestinal segment as a result of peristalsis, and in many cases cause intestinal obstruction. Its etiology can be benign, malignant or idiopathic, mostly given by different entities, including intestinal diverticula, adhesion bands, vascular malformations, neoplasms, among others. The vast majority occur in paediatric patients, however, up to 5% of these are documented in adult patients and their main etiology in this age group is neoplasms. We present a case of intestinal intussusception secondary to Burkitt's lymphoma that received management at the San Ignacio University Hospital in Bogotá, Colombia.

Case presentation: This case report describes the clinical presentation and diagnostic approach of a patient who presented to the Emergency Department (ED) with clinical symptoms suggestive of intestinal obstruction, in which imagological findings of intestinal intussusception are documented, and led to an exploratory laparotomy where the presence of a jejunal-jejunal intussusception conditioned by a neoplastic mass is confirmed. This mass was resected and sent for histopathological study, where the diagnosis of Burkitt lymphoma with intestinal involvement confirmed. Subsequently, the patient had a good clinical evolution and is discharged to continue his long-term follow-up by the oncology service.

Conclusion: Intestinal obstruction in adults secondary to intussusception is rare and case reports in which the etiology is Burkitt Lymphoma are scarce. Likewise, due to the unspecific symptoms, the surgeon must have the clinical suspicion to be able to carry out a successful diagnostic approach. This case shows how the use of appropriate diagnostic images can guide diagnosis and direct the appropriate surgical management of this surgical emergency.

Keywords: Intussusception; Burkitt's lymphoma; Intestinal obstruction

Introduction

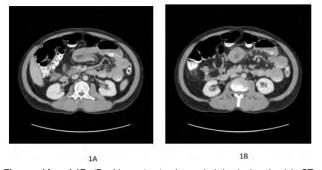
A 53-year-old male patient from Bogotá, Colombia, who presented to the ED of the Hospital Universitario San Ignacio because of 15 days of right upper quadrant abdominal pain of low intensity which worsened 4 days before the hospital admission; it was accompanied by vomiting and absence of bowel movement. Physical examination revealed normal vital signs, no cardiopulmonary auscultatory altered findings. Positive bowel sounds and mild periumbilical pain with palpation and no signs of peritoneal irritation were found on abdominal examination. Neither have we palpated masses nor abdominal wall defects. We carried out laboratory tests including complete blood counts, serum creatinine, liver function tests and electrolytes; all values were normal. And also, radiological studies were requested, first and abdominal ultrasound was carried out, which reported enlarged lymph nodes in the hepatic hilum. With this finding a double contrast enhanced abdominal and pelvic Computed Tomography (CT) was requested.

Case Presentation

On the CT the radiologist reported thickening of the jejunal walls with invagination of a small bowel segment into an adjacent segment on the left flank with a target sign appearance, and dilatation of proximal small bowel loops (Figures 1A and 1B); with these findings a jejunojejunal intussusception with signs of partial mechanical small bowel obstruction was suggested. In addition, three masses of solid attenuation adjacent to the hepatic hilum, vesicular fossa and II liver segment were depicted. Also, interaortocaval and coeliac enlarged lymph nodes and an eccentric filling defect of the main portal vein suggestive of thrombosis were reported (Figure 2). With these findings the patient was programmed for an exploratory laparotomy, 100 cm apart from

J Clin Case Rep, an open access journal ISSN: 2165-7920

the Treitz ligament a 4 cm rounded mass dependent of the jejunum configuring a jejuno-jejunal intussusception (Figure 3) with dilated proximal jejunum bowels and collapsed distal bowels. A segmentary



Figures 1A and 1B: (Double contrast enhanced abdominal and pelvic CT. Venous phase, axial reconstruction); Wall thickening of left flank small bowel loop which is telescoped into another adjacent small bowel loop with its mesentery, configuring a target sign (hyperdense-hypodense-hyperdense appearance).

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Received December 01, 2019; Accepted December 09, 2019; Published December 16, 2019

Citation: Ustariz M, Carlos A, Boscan S, Sophia C, Lesmes P, et al. (2019) Intestinal Intussusception Secondary to Burkitt's Lymphoma in an Adult: A First Case Report in Latin America. J Clin Case Rep 9: 1302.

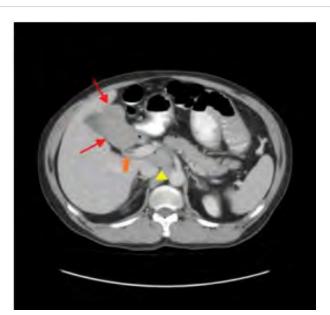


Figure 2: (Double contrast enhanced abdominal and pelvic CT. Venous phase, axial reconstruction); Two contiguous oval masses adjacent to hepatic hilum and vesicular fossa, of homogenous low density (compared to the liver. Red arrow) and interaortocaval and coeliac enlarged lymph nodes (arrowhead); Eccentric filling defect of the main portal vein (partially seen. Orange arrow).

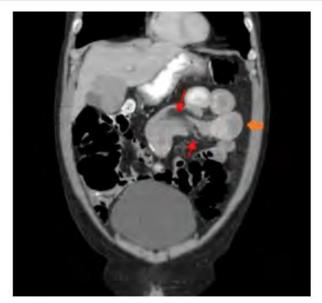


Figure 3: (Double contrast enhanced abdominal and pelvic CT. Venous phase, coronal reconstruction); Intussuscipiens and intussuscepted small bowel loop on the left flank with mesentery fat and vessels (red arrow) in between the bowel's walls associated with proximal bowel dilatation (partially seen, orange arrow).

intestinal resection was made and a termino-terminal jejuno-jejunal anastomosis with a mechanical suture was performed. Afterwards, a systematic abdominal cavity revision was made, only finding a 4.0 \times 5.0 cm multilobulated mass near the hepatic hilum, adjacent to the medial aspect of the gallbladder and in contact to the IV and V hepatic segments; multiple fragments were obtained for histopathological

studies. Reports of both biopsies confirmed the presence of a Highgrade B cell NonHodgkin Lymphoma with a germinal centre variant, positive for CD20, C45, CD10, Bc16 and C-Myc (90%) tumoral markers and a 100% Ki-67. All of which are diagnostic findings for Burkitt's lymphoma.

Discussion

The first case of intestinal intussusception was reported by Bardette, et al., in 1674 and the first description of its surgical management was made by Sir Jonathan Hutchinson in 1871 [1]. Intestinal intussusceptions are often present in paediatric patients presenting up to 95% of the cases, only the 5% left are diagnosed in adults [1,2]. Its causes can be varied, however, in adults they are frequently associated with processes of neoplastic origin and up to 20% can be of idiopathic origin. On the other hand, Burkitt's Lymphoma (BL) is an aggressive neoplasm derived from B cells, occurs between the third and fourth decade of life and has an incidence close to 0.6 per million, there are 4 variants:

- 1. Sporadic
- 2. Endemic
- 3. Associated with HIV
- 4. Associated with post-transplant lymphoproliferative syndrome.

Multiple cases of intestinal intussusception due to BL have been reported. One of the most extensive reviews by is done by Prabin et al. In spite of documenting a significant number of cases, the majority were in paediatric patients [3]. In fact, to date less than 20 cases have been reported in the adult population. One of these by Simson et al., Who in 2017 reported the first case in the United Kingdom, reported the intussusception in a 22-year-old patient whose diagnosis was made by computed tomography of the abdomen and subsequently led to surgical management [4]. There are also reports made in Turkey, Portugal and Korea, among others, without having described any in Latin America so far [5-7]. Intestinal intussusception should be considered a surgical emergency and its diagnosis should not be delayed. Despite this, its clinical picture is usually nonspecific and resembles that of an intestinal obstruction of other etiologist, which makes it a diagnostic challenge; the most frequent symptoms being abdominal pain (94%), nausea (76%), emesis (65%), diarrhoea (14%) and blood in stool (7%) [8].

Imaging findings, specifically in computed tomography of the abdomen, are highly suggestive. It is typically presented as a targetshaped or "sausage", depending on the axial projection [9,10] where the afferent bowel, fat, mesenteric vessels, the efferent loop and intraluminal space can be identified within the mass. The image of a bowel inside a bowel with or without mesenteric fat or vessels is pathognomonic. Also, edema of the intestinal walls can be found due to alteration in blood flow, with thickening of the intestinal walls, which makes it difficult to differentiate a mass as a starting point for intussusception. It can be associated with proximal intestinal loops to the intussusception site [9].

In terms of management, this may vary between age groups. While in paediatric patients the causes are usually "benign" and medical management can be considered, in adult patients as it is commonly associated with neoplastic diseases it usually requires surgery [5] so the indication is surgical management, and usually segmental bowel resections are required that may or may not include the masses that condition the intussusception; in case of resecting it, it must be done

Page 2 of 3

ISSN: 2165-7920

Page 3 of 3

with oncological principles [2]. After receiving surgical management, patients should continue in multidisciplinary follow-up by the oncology and haematology services to define the long-term management of the underlying disease.

Conclusion

Intestinal intussusceptions secondary to Burkitt's lymphoma are an extremely rare condition that requires early diagnosis and timely management. This case shows how the appropriate use of diagnostic images ensures an early preoperative diagnosis and the surgical management of the patient which represent satisfactory surgical outcomes and successful postoperative evolution.

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