

Intestinal intussusception due to Burkitt's lymphoma in an adult. First report in Latin America

Intususcepción intestinal por linfoma de Burkitt en un paciente adulto. Primer reporte en Latinoamérica

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Abstract

Intestinal intussusception is a pathology in which an intestinal segment and its mesentery are telescoped into an adjacent intestinal segment as a result of peristalsis, and in many cases cause intestinal obstruction. Its etiology can be variable, including intestinal diverticula, adhesion bands, vascular malformations, neoplasms, among others. The vast majority occur in pediatric patients, however, up to 5% of these are documented in adult patients and their main etiology in this age group are neoplasms. We present a case of intestinal intussusception secondary to Burkitt's lymphoma that received management at fourth level hospital in Bogotá, Colombia.

Keywords: Intussusception. Burkitt's lymphoma. Bowel obstruction. Lymphoma.

Resumen

La intususcepción intestinal es una patología en la que un segmento intestinal y su mesenterio se invaginan al segmento intestinal contiguo. Su etiología puede ser benigna, maligna o idiopática, dentro de las que se incluyen divertículos intestinales, bandas adherenciales, malformaciones vasculares y neoplasias, entre otras. La mayoría se presentan en pacientes pediátricos, pero hasta un 5% se documentan en pacientes adultos y su principal etiología en este grupo etario son las neoplasias. Presentamos un caso de intususcepción intestinal secundaria a linfoma de Burkitt que recibió manejo en un hospital de cuarto nivel en Bogotá, Colombia.

Palabras clave: Intususcepción. Linfoma de Burkitt. Obstrucción intestinal. Linfoma.

Introduction

Intestinal intussusceptions in adult patients are rare. They appear with the typical clinical symptoms of an intestinal obstruction and are frequently associated with neoplastic processes of different types. Those associated with Burkitt's lymphomas are extremely rare and the cases reported in the world literature are few.

Clinical case

53-year-old male, from Bogota, Colombia, who consulted with the emergency service at San Ignacio University Hospital because of clinical features of 15 days of progress which consisted of abdominal pain localized in the right hypochondrium, of low intensity, and then aggravated 4 days previous to the consultation.

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It was combined with multiple emetic episodes and absence of bowel movement. The patient denied any other associated symptom. At the physical examination, the vital signs were normal, with no alterations at the cardiopulmonary auscultation. The abdominal evaluation showed the presence of bowel sounds, with mild periumbilical tenderness and no signs of peritoneal irritation. No hernial defect was shown and no mass was felt.

Among the paraclinical tests requested, the hemogram showed no alterations at the cellular lines, the hemoglobin was normal, and the platelet count was within the normal ranges; normal creatinine and urea nitrogen values; normal hepatic profile and electrolytes. Additionally, imaging studies were requested. First, a complete abdomen ultrasound scan, in which a lymph nodal mass in relation to the hilum of the liver was registered, without any other pathological finding. Considering these findings, it was decided to request a CT scan of the abdomen and pelvis with intravenous and oral contrast dye, where it was found a thickening at the jejunum walls with invagination of an intestinal loop into the interior of a contiguous loop, at the left flank, with target sign morphology (Figs. 1 and 2). These findings suggested an intestinal jejunum-jejunal intussusception and concomitant signs of partial mechanical intestinal obstruction due to the dilation of the loops proximal to the intussusception site. Besides, there was evidence of two apparently solid masses adjacent to the gallbladder fossa and the hilum of the liver (7.3 x 3.9 cm) and another one adjacent to segment II of the liver (4.3 x 3.1 cm), with presence of inter aorto cava adenomegalies and adjacent to the celiac artery, and a defect of partial filling and eccentric localization of the main portal vein, suggesting partial thrombosis (Fig. 3).

Due to these findings, it was decided to perform an exploratory laparotomy that displayed, at 100 cm from the ligament of Treitz, a round mass dependent on the jejunum, of approximately 4 cm, that lead a jejunum-jejunal intussusception (Fig. 4) with dilated jejunum proximal loops and the distal ones collapsed. An intestinal segmental resection with mechanical suture and a jejunum-jejunal end-to-end anastomosis with mechanical suture were performed. Immediately, a systematic revision of the abdominal cavity was carried out, finding a mass at the hilum of the liver, in relation to the gallbladder medial border, of approximately 4 x 5 cm, multi-lobal, in contact with segments IV and V of the liver. Samples of this mass were taken for a histopathological examination.

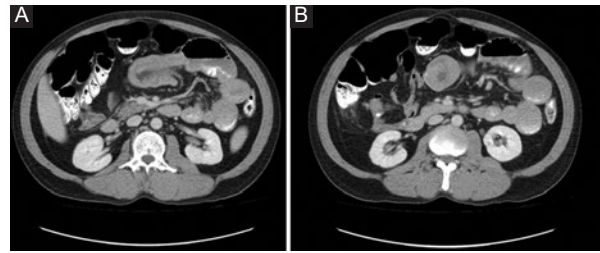


Figure 1. CT scan of the abdomen with oral and intravenous contrast dye, venous phase, axial section. **A:** at the left flank, a thickening of the intestinal loop walls, with invagination of a thin loop inside of another contiguous thin loop is observed. **B:** target sign.

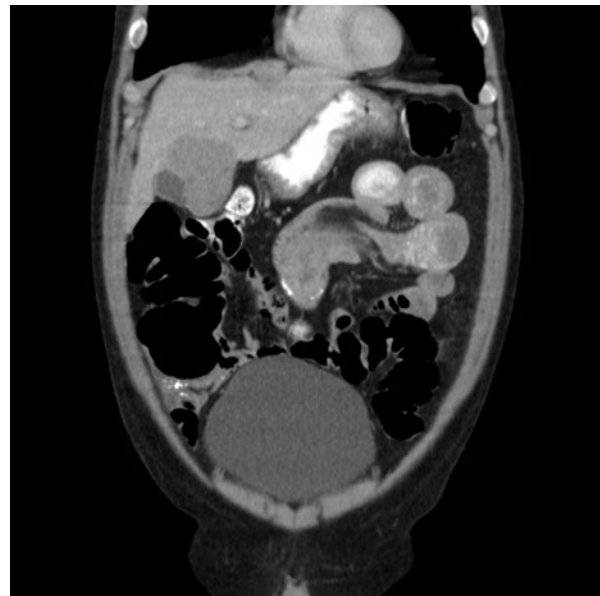


Figure 2. CT scan of the abdomen with oral and intravenous contrast dye, venous phase, coronal section, in which an intussusceptum thin loop and the intussusciens loop are observed, together with vessels and mesenteric fat among them, associated with dilation of the thin proximal intestinal loops.

The histopathological reports of both samples confirmed the presence of a non-Hodgkin B-cell lymphoma of high-grade, germinal center variant, positive to tumor markers CD20, CD45, CD10, Bc16 and C-myc (90%), with Ki-67 of 100%. Said findings established the diagnose of Burkitt's lymphoma.

Discussion

The first case of intestinal intussusception was reported by Bardette in 1674, and the first description of its surgical management was written in 1871 by Sir Jonathan Hutchinson¹. Even though intestinal intussusceptions appear mainly in pediatric patients, reaching up

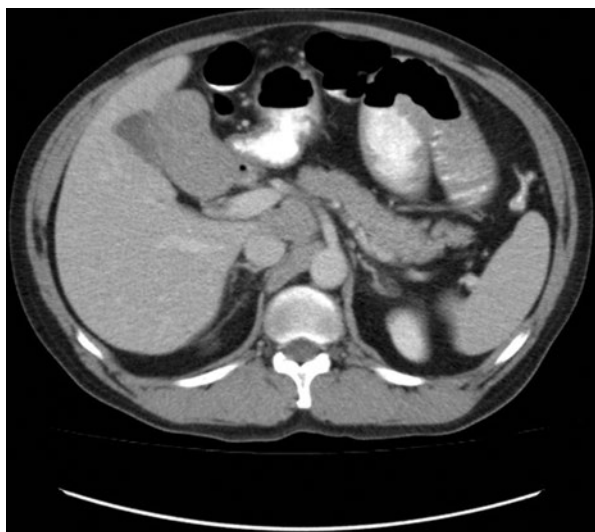


Figure 3. CT scan of the abdomen with oral and intravenous contrast dye, venous phase, axial section, in which two hypodense, contiguous, oval, and seemingly solid masses, adjacent to the gallbladder fossa and the hilum of the liver, as well as inter aorto cava adenomegalies, adjacent to the celiac artery, are observed. There was also an eccentric filling defect of the main portal vein, suggesting thrombosis (not displayed).

to a 95% of cases, the other 5% occurs in adults^{1,2}. The causes may vary; however, in adults, it is frequently associated to processes of neoplastic origin and up to a 20% may be of idiopathic origin. As regards Burkitt's lymphoma, it is an aggressive neoplasm derived from B-cells, which occurs between the third and fourth life decades, has an incidence near to 0.6 per million and of which four variants exist: sporadic, endemic, associated to the human immunodeficiency virus, and associated to the post-transplant lymphoproliferative syndrome².

There have been multiple cases reported describing intestinal intussusception due to Burkitt's lymphoma. In one of the most extensive revisions, carried out by Sharma et al.³, even though an important number of cases were documented, most of them concerned pediatric patients. In fact, up to date, less than 20 cases have been reported in adults. Simson et al.⁴, in 2017, reported the first case in the United Kingdom of a 22-year-old patient whose diagnose was performed through a CT scan of the abdomen and was later derived to surgical management. Reports have also been carried out in Turkey⁵, Portugal⁶ and Korea⁷, among others, but so far none has been described in Latin America.

Intestinal intussusception must be considered a surgical emergency and its diagnose should not be delayed.



Figure 4. Intussuscepted jejunum loop.

Notwithstanding, clinical features are usually non-specific and resemble those of an intestinal obstruction of other etiologies, turning it into a diagnostic challenge. Most frequent symptoms are abdominal pain (94%), nausea (76%), emesis (65%), diarrhea (14%), and blood in the feces (7%)⁸.

Imaging findings, specifically in the CT scan of the abdomen, are very suggestive. It tends to occur as a mass that resembles the sausage and target signs, depending on the axial section^{9,10}, where the afferent loop, fat and the mesenteric vessels inside the mass, the efferent loop, and the intraluminal space can be identified. The configuration of loop inside of loop is pathognomonic, with or without fat and mesenteric vessels. It can also be found that there is edema of the intestinal walls due to an abnormal blood flow, with thickening of the loop walls. This makes it difficult to differentiate a mass as the starting point of the intussusception. It may be associated with a proximal intestinal obstruction, and in such cases a dilation of the intestinal loops proximal to the point of the intussusception is observed⁹.

As regards management, it can vary according to the age group. Whereas in pediatric patients the causes are usually "benign" and medical management might

be considered, in adults, as it tends to be associated to neoplastic diseases, a definitive management is required⁵, thus surgery is indicated. Usually, segmental resections of the intestine are required, which must include the masses that condition the intussusception; and in case of resection, it must be performed with oncology principles².

After surgical management, patients must continue with a multidisciplinary follow up from the oncology and hemato-oncology services in order to determine a long term treatment for the underlying disease.

Conclusions

The intestinal intussusception secondary to Burkitt's lymphoma is an extremely rare condition that requires an early diagnose and timely management. This case portraits the appropriate use of imaging to perform an early preoperative diagnosis and plan the surgical management of the patient, obtaining good progress and a successful postoperative outcome.

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Conflicts of interest

The authors state they have no conflicts of interest.

Ethical disclosures

Protection of people and animals. The authors declare that no experiment was performed in human beings nor in animals for this investigation.

Data confidentiality. The authors declare that they have followed their center's protocols on the publication of patient data.

Right to privacy and informed consent. The authors have obtained the informed consent of the patients and/or subjects referred to in the article. This document is in the possession of the corresponding author.

References

1. Yalamarthy S, Smith RC. Adult intussusception: case reports and review of literature. *Postgrad Med J*. 2005;81:174-8.
2. Ozant A. Adult multicentric Burkitt lymphoma with bowel obstruction due to intussusception. *Turk J Gastroenterol*. 2018;2018:361-4.
3. Sharma P, Balasingham S, Stawiariski K, Xu M. Burkitt lymphoma as a lead point for jejunojejunal intussusception in a human immunodeficiency virus patient. *Clin J Gastroenterol*. 2017;10:342-50.
4. Simson R, Planner A, Alexander R. Adult Burkitt's lymphoma presenting as intussusception: first UK case report. *Ann R Coll Surg Engl*. 2017;99:e206-8.
5. Yagmur Y, Gumus S. Burkitt's lymphoma causing intussusception in adults : report of two cases and review of the literature. *J Gastroenterol Hepatol Res*. 2015;4:1702-6.
6. Fernandes C, Pinho R, Ribeiro I, Silva J, Ponte A, Leite S, et al. Recurrent cecocolic intussusception in a young woman. *Gastroenterol Hepatol*. 2015;38:586-8.
7. Baek D, Heo D, Oh S, Hwang J, Hwang J, Park HS, et al. A case of jejunal intussusception caused by Burkitt lymphoma in an acquired immunodeficiency syndrome patient. *Infect Chemother*. 2018;50:51-4.
8. Castro Medina CA, Jiménez HC. Clinical case presentation: diagnosis and treatment of idiopathic intussusception in adults. *Rev Colomb Gastroenterol*. 2015;30:474-8.
9. Kim YH, Blake MA, Harisinghani MG, Archer-Arroyo K, Hahn PF, Pitman MB, et al. Adult intestinal intussusception: CT appearances and identification of a causative lead point. *Radiographics*. 2006;26:733-45.
10. Felix EL, Cohen MH, Schwartz JH. Adult intussusception; case report of recurrent intussusception and review of the literature. *Am J Surg*. 1974;131:758-61.