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Ileocolic Intussusception Due to a Gastrointestinal Stromal Tumor: A Case Report

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Abstract | **Case Report**

Adult intussusception is a rare entity which is distinct from paediatric cases in incidence, The overall incidence of intussusception in adulthood has been estimated to be around 2–3 cases/1,000,000 population/year. Ileocolic intussusception in adults is a unique variant in which nearly 100% of cases have a malignant lead point. In our report we present a case of ileocolic intussusception in a 60-year-old female patient caused by a GIST located in the terminal ileum, acting as the apex of intussusception. The female patient was admitted to hospital for abdominal pain, vomiting, nausea, and inability to defecate. The result of CT scan of the abdomen and pelvis showed ileal intussusception, which was confirmed peroperatively. Bowel resection was performed. The final pathologic diagnosis indicated the gastrointestinal stromal tumor of the ileum. The diagnosis of intussusception in adults is delicate, and timely surgical treatment can be vital. Patients with the palpable abdominal mass, digestive tract obstruction, gastrointestinal bleeding, or lead point computed tomography must undergo a surgical examination. Given a high risk of malignancy, primary surgical resection using oncologic principles presents the best option for the treatment of ileocecal intussusception in adults.

Keywords: Iléal intussusception, GIST tumors.

INTRODUCTION

Intussusception is defined as the invagination of a part of the gastrointestinal tract into an adjacent part, The overall incidence of intussusception in adulthood has been estimated to be around 2–3 cases/1,000,000 population/year [1]. Their distribution in the small intestine indicates that 17.7% are in the duodenum, 47.6% in the jejunum, and 34.7% in the ileum [2, 3]. In 60% of cases, intussusception of the small intestine in adults is caused by benign lesions. The rest are caused by malignancy (30%) or are idiopathic (10%). However, most colonic intussusceptions are caused by malignancy (60%) (4). We present an unusual case of ileocolic intussusception in a 60-year-old female patient caused by a GIST located in the terminal ileum, acting as the apex of intussusception.

CASE REPORT

A 60-year-old woman presented to our hospital with abdominal pain, vomiting, nausea, and inability to defecate. His medical history included a type-2-diabetes. Lab tests were at normal ranges. An abdominal Xray revealed small bowel obstruction with marked small bowel air–fluid levels. The additionally performed CT-scan showed typical signs of an ileocolic invagination (the terminal ileum telescoping into the right colon). At laparotomy. The small bowel was dilated, and the ileum was invaginated into the colon (Figure 1). The ileum was reduced but at the cost of an ileal perforation wich gave way to the intraluminal tumor (Figure 2). Ileal resection that included the tumor and the perforation, and entero-enteric anastomosis were performed because the mass was possibly malignant (figure3).

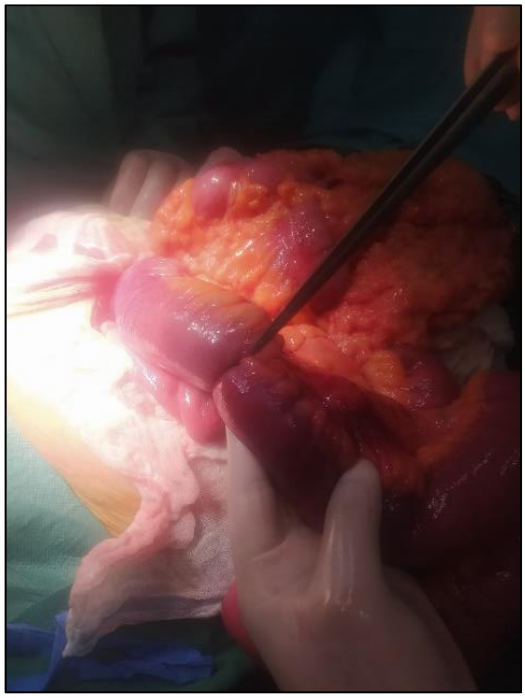

Figure 1: Per operative view showing bowel Intussusception

Figure 2: per operative view showing the tumor causing the Intussusception after reduction

Figure 3: ileal resection piece

The pathology report confirmed that the neoplasm was a small bowel GIST. The margins of surgical resection and all identified mesenteric lymph nodes were negative for malignancy. The patient is under surveillance

DISSCUSION

Intussusception was first described by Dr. Paul Barbette in 1674 and first treated by Sir Jonathan Hutchinson in 1871 (5). Intussusception in adults is a very rare occurrence. It represents 5% of all intussusceptions and is the cause of 1% of all intestinal obstructions, 0,08% of all abdominal surgeries and 0,003-0,02% of all hospital admissions [4].

Typical locations for bowel intussusception in the gastrointestinal tract are junctions between freely moving bowel segments and retroperitoneally or adhesionally fixed segments. According to the involved bowel segments, intussusceptions can be classified as entero-enteric, ileo-colic, colo-colic, colo-rectal or recto-rectal [6,7].

In direct contrast to pediatric etiologies, adult intussusception is associated with an identifiable cause in 90% of symptomatic cases, with an idiopathic cause in 10% of cases [15]. Benign or malignant neoplasms cause two-thirds of cases of intussusception in adults; the remaining cases are caused by infections, postoperative adhesions, Crohn granulomas, intestinal ulcers (Yersinia), and congenital abnormalities such as Meckel diverticulum. In our case, the patient's past medical history was negative for gastrointestinal diseases. Also, our patient had no previous abdominal surgery or abdominal trauma. Of the cases caused by neoplasms, 50% of them are malignant [16].

Malignant lesions include primary tumors such as carcinoids, adenocarcinoma, malignant polyps, GISTs, leiomyosarcomas, lymphoma and metastatic tumors, most commonly melanoma [17]. The most common malignant cause of colonic intussusception is primary colonic adenocarcinoma and the most common benign cause is colonic lipoma [16]. Ileocolic intussusception in adults is a unique variant in which nearly 100% of cases have a malignant lead point, namely, cecal adenocarcinoma involving the ileocecal valve [18].

Small bowel GISTs are usually asymptomatic, especially in their early stages and they often go unrecognized until severe symptoms ensue, which can create surgical emergencies [8,10]. Although slow-growing, GISTs can grow very large before producing signs and symptoms, as they tend to displace adjacent structures without invasion [11]. In addition, they can spread to the liver, lungs, and bones via the bloodstream, bypassing the local lymph nodes [12]. They are often detected incidentally on physical examination, radiologic imaging, endoscopy, or laparotomy, but eventually the majority of patients develop symptoms because of disease progression [13].

Symptomatic GISTs often present with non-specific and vague abdominal symptoms and signs [14] The most common clinical findings include an abdominal mass, pain, bleeding, weight loss, nausea, vomiting and obstructive ileus [8,11]. These symptoms mainly depend on the size and the location of the tumor, with lesions distal to the ligament of Treitz having a tendency to present with either obstruction or bleeding [19]. GISTs tend to grow in an extraluminal fashion; however, they can also erode into the lumen of the gastrointestinal tract inducing significant hemorrhage or anemia from occult bleeding [8]. They can also rupture into the peritoneal cavity causing significant hemorrhage [20]. In addition to symptoms from mass effect or bleeding, GISTs can cause intussusception or rarely intestinal obstruction [21].

In this case the patient was asymptomatic until intestinal obstruction developed. Of note is that a rare tumor acted as a lead point for the ileocolic intussusception, which is also a very uncommon condition. Intussusception accounts for only 1% to 5% of all cases of intestinal obstruction in adults and is rarely diagnosed preoperatively [22,23]. This is mainly related to the paucity of patients and the non-specific complaints and physical findings of intussusception that can be confused with other causes of intestinal obstruction. Common physical findings include abdominal distension and tenderness, an abdominal mass, colicky pain, nausea, vomiting, change in bowel habits, constipation, hypoactive to absent bowel sounds, and bleeding [24]. The classic triad of abdominal mass, tenderness, and haemoglobin-positive stools is rarely found [25].

Because of the non-diagnostic physical findings of intussusception, most patients undergo further investigation with various imaging modalities. An ideal diagnostic algorithm has to be defined; however, CT scanning has been reported to be the most useful tool for the diagnosis of intestinal intussusception, and it appears to be superior to other contrast studies, ultrasonography, or endoscopy [26, 27]. Furthermore, as the majority of adult intussusception is caused by an underling neoplastic lesion, abdominal CT should probably be the first imaging investigation upon suspicion of intussusceptions, and can provide additional staging information. The density of the intussusceptum within the lumen of the intussuscipien gives the characteristic "target sign" or "sausage shaped appearance", [25].

Surgical resection is recommended in nearly all cases of adult intussusception, because of the high prevalence of structural anomalies and the relatively high risk of underlying malignancy. However, the issue of reduction versus mandatory primary resection

remains a topic of some controversy. Weilbacher and associates [28] established the principle of mandatory primary resection without reduction, because of the high incidence of underlying malignancy. They also claimed that reduction includes the theoretical risk of intraluminal seeding or venous embolization in regions of ulcerated mucosa [28]. On the other hand, mandatory resection necessitates the excision of a long segment of small bowel, which may compromise the mesenteric vessels. Therefore it has been proposed that gentle operative reduction, when feasible, can be attempted safely before resection, to avoid the unnecessary excision of a healthy bowel [29].

CONCLUSION

This case presents an unusual malignant cause of adult intussusception and highlights the importance of computed tomography scanning in the accurate diagnosis of this rare entity.

REFERENCES

1. Manouras, A., Lagoudianakis, E. E., Dardamanis, D., Tsekouras, D. K., Markogiannakis, H., Genetzakis, M., ... & Katergiannakis, V. (2007). Lipoma induced jejunojejunal intussusception. *World journal of gastroenterology: WJG*, *13*(26), 3641.
2. Weiss, N. S., & Yang, C. P. (1987). Incidence of histologic types of cancer of the small intestine. *Journal of the National Cancer Institute*, *78*(4), 653-656.
3. Lewis, J. J., & Brennan, M. F. (1996). Soft tissue sarcomas. *Current problems in surgery*, *33*(10), 820-872.
4. Potts, J., Al Samaraee, A., & El-Hakeem, A. (2014). Small bowel intussusception in adults. *The Annals of The Royal College of Surgeons of England*, *96*(1), 11-14.
5. Hutchinson, J. (1874). A successful case of abdominal section for intussusception, with remarks on this and other methods of treatment. *Medico-chirurgical transactions*, *57*, 31.
6. Zubaidi, A., Al-Saif, F., & Silverman, R. (2006). Adult intussusception: a retrospective review. *Diseases of the colon & rectum*, *49*, 1546-1551.
7. Marinis, A., Yiallourou, A., Samanides, L., Dafnios, N., Anastasopoulos, G., Vassiliou, I., & Theodosopoulos, T. (2009). Intussusception of the bowel in adults: a review. *World journal of gastroenterology: WJG*, *15*(4), 407.
8. Crosby, J. A., Catton, C. N., Davis, A., Couture, J., O'Sullivan, B., Kandel, R., & Swallow, C. J. (2001). Malignant gastrointestinal stromal tumors of the small intestine: a review of 50 cases from a prospective database. *Annals of surgical oncology*, *8*, 50-59.
9. Nagorney, D. M., Sarr, M. G., & McIlrath, D. C. (1981). Surgical management of intussusception in the adult. *Annals of surgery*, *193*(2), 230.
10. Mahvi, D.M., & Stone, J. (2000). Small Bowel. Clinical Oncology. Edited by: Abeloff MD, Armitage JO, Lichter AS, et al. New York: *Churchill Livingstone*, *1586*-1610.
11. Ludwig, D. J., & Traverso, L. W. (1997). Gut stromal tumors and their clinical behavior. *The American journal of surgery*, *173*(5), 390-394.
12. Tworek, J. A., Appelman, H. D., Singleton, T. P., & Greenson, J. K. (1997). Stromal tumors of the jejunum and ileum. *Modern pathology: an official journal of the United States and Canadian Academy of Pathology, Inc*, *10*(3), 200-209.
13. Kindblom, L.G. (2003). Education Session E450, oral presentation "Gastrointestinal Stromal Tumours Diagnosis, Epidemiology and Prognosis" in Gastrointestinal Stromal Tumour: Current management and Future Challenges. Available from http://www.asco.org/ASCO/Abstracts+%26+Virtual+Meeting/Speaker?&spk=Kindblom%2C+Lars-Gunnar+%5Bfau%5D. 2003, Chair: Blanke CD. ASCO, last accessed on December 9, 2008, http://media.asco.org/asco/meetings_education/module/audio/frame.asp?EventName=vm2003&ID=875&mediaURL=/media&ServerName=media.asco.org
14. Strickland, L., Letson, G. D., & Muro-Cacho, C. A. (2001). Gastrointestinal stromal tumors. *Cancer control*, *8*(3), 252-261.
15. Willekens, I., Vandenbroucke, F., Van Nieuwenhove, Y., Hoorens, A., & De Mey, J. (2008). Ileocolic invagination as a complication of a cecal adenocarcinoma. *Journal of Radiology Case Reports*, *2*(5), 29.
16. Namikawa, T., Okamoto, K., Okabayashi, T., Kumon, M., Kobayashi, M., & Hanazaki, K. (2012). Adult intussusception with cecal adenocarcinoma: Successful treatment by laparoscopy-assisted surgery following preoperative reduction. *World Journal of Gastrointestinal Surgery*, *4*(5), 131.
17. Verre, L., Rossi, R., Gaggelli, I., Piccolomini, A., Podzemny, V., & Tirone, A. (2012). Ileocecal-colonic intussusception caused by cecal adenocarcinoma A case report. *Annali Italiani di Chirurgia*, *2012*, S2239253X12019081-S2239253X12019081.
18. Cheung, W. K., Tu, C. C., & Wang, K. L. (2008). Ileocolic intussusception secondary to cecal adenocarcinoma. *Journal of the American Geriatrics Society*, *56*(11), 2148-2150.
19. Lewis, B. S. (1994). Small intestinal bleeding. *Gastroenterology Clinics of North America*, *23*(1), 67-91.
20. Hirasaki, S., Fujita, K., Matsubara, M., Kanzaki, H., Yamane, H., Okuda, M., ... & Saeki, H. (2008). A ruptured large extraluminal ileal gastrointestinal stromal tumor causing hemoperitoneum. *World Journal of Gastroenterology: WJG*, *14*(18), 2928.

21. Pidhorecky, I., Cheney, R. T., Kraybill, W. G., & Gibbs, J. F. (2000). Gastrointestinal stromal tumors: current diagnosis, biologic behavior, and management. *Annals of Surgical Oncology*, *7*, 705-712.
22. Eisen, L. K., Cunningham, J. D., & Aufses Jr, A. H. (1999). Intussusception in adults: institutional review. *Journal of the American College of Surgeons*, *188*(4), 390-395.
23. Begos, D. G., Sandor, A., & Modlin, I. M. (1997). The diagnosis and management of adult intussusception. *The American Journal of Surgery*, *173*(2), 88-94.
24. Wang, L. T., Wu, C. C., Yu, J. C., Hsiao, C. W., Hsu, C. C., & Jao, S. W. (2007). Clinical entity and treatment strategies for adult intussusceptions: 20 years-experience. *Diseases of the colon & rectum*, *50*, 1941-1949.
25. Azar, T., & Berger, D. L. (1997). Adult intussusception. *Annals of surgery*, *226*(2), 134.
26. Takeuchi, K., Tsuzuki, Y., Ando, T., Sekihara, M., Hara, T., Kori, T., & Kuwano, H. (2003). The diagnosis and treatment of adult intussusception. *Journal of clinical gastroenterology*, *36*(1), 18-21.
27. Tan, K. Y., Tan, S. M., Tan, A. G., Chen, C. Y., Chng, H. C., & Hoe, M. N. (2003). Adult intussusception: experience in Singapore. *ANZ journal of surgery*, *73*(12), 1044-1047.
28. David, W., John, A. B., David, H., & William Ogden, I. I. (1971). Intussusception in adults: Review of 160 cases. *Am J Surg*, *121*, 531-535.
29. Baig, M. K., Hussain, S., Wise, M., & Wexner, S. D. (2000). Controversy in the treatment of adult long ileocolic intussusception: case report. *The American Surgeon*, *66*(8), 742-743.