Published online 2014 December 20.

Case Report

Jejunojejunal Intussusception Caused by a Jejunal Villous Adenoma Polyp in an Adult

Seyed Mohammad Hossein Kashfi¹; Faegheh Behboudi Farahbakhsh¹; Mina Golmohammadi¹; Ehsan Nazemalhosseini Mojarad²; Pedram Azimzadeh²; Shahrokh Iravani³; Hamid Asadzadeh Aghdaei^{1,*}; Katayoun Gohari Moghaddam³; Mohammadreza Zali²

¹Basic and Molecular Epidemiology of Gastroenterology Disorders Research Center, Shahid Beheshti University of Medical Sciences, Tehran, IR Iran ²Gastroenterology and Liver Diseases Research Center, Shahid Beheshti University of Medical Sciences, Tehran, IR Iran

³Aja Cancer Research Center (ACRC), Aja University of Medical Sciences, Tehran, IR Iran

*Corresponding author: Hamid Asadzadeh Aghdaei, Basic and Molecular Epidemiology of Gastrointestinal Disorders Research Center, Shahid Beheshti University of Medical Sciences, Tehran, IR Iran. Tel: +98-22432515, E-mail: hamid.assadzadeh@gmail.com

Received: November 17, 2014; Revised: November 30, 2014; Accepted: December 15, 2014

Introduction: Intussusception is telescoping or invagination of one part of the intestine (intussusception) into an adjacent section (intussuscipiens) and it may present as a life threatening condition. Intussusception is so rare in adults and small intestine intussusception in adults is usually due to benign entity; whereas in large intestine, malignant neoplasms can be the etiology. Here, we reported a rare case of adult jejunojejunal intussusception due to a jejunal villous adenoma polyp in an Iranian female patient.

Case Presentation: A 33-year female Iranian patient referred to emergency department in Gastroenterology and liver diseases Research Center, Shahid Beheshti University of Medical Sciences with extreme abdominal pain, nausea and constipation. Plain abdominal computed tomography (CT) scan revealed a mass suggesting intussusception in the jejunum. Balloon association enteroscopy showed two 10 mm and 40 mm polyps in jejunum. The polyp was a villous adenoma confirmed by pathology. Exploratory laparotomy revealed jejunojejunal intussusceptions and exploratory laparotomy with jejunoileal anastomosis was performed.

Conclusions: Nonspecific symptoms of an adult intussusception might result in a late diagnosis and imaging modalities including CT scan play a significant role in the diagnosis. Surgical operation is the best treatment in adult intussusception leading to the best results.

Keywords:Intussusception; Surgery; Villous; Tomography

1. Introduction

Intussusception is telescoping or invagination of one part of the gastrointestinal tract (intussusception) into an adjacent section (intussuscipiens) and it may present as a life threatening condition. Intussusception mostly occurs in childhood and is rare in adults. It has been reported that the incidence of intussusceptions in adults is approximately 5% and this phenomenon is the leading cause of 1% of all bowel obstruction in adults (1, 2). Small intestine intussusception in adults usually arises from benign entity; however, when it involves the large intestine malignant neoplasms should be considered (3). The specific symptoms of intussusception in adults are chronic colicky pain and partial intestinal obstruction (4). However, nausea, abdominal pain, and vomiting, constipation, weight loss and fever are other presentations (4). In contrast to childhood intussusception which is idiopathic in 90% and only 10% of them have a pathological etiology, in 90% of adult cases there is a specific pathological lead point, from which 65% raised by a polyp, a neoplastic mass or submucosal lipoma (3). As most adult patients with intussusception have an obvious pathology entity, most of them require surgical resection. There is a controversy about the extent of surgical resection and reduction before excision (5). Here in this study, we reported a rare case of adult jejunojejunal intussusception due to a jejunal villous adenoma polyp in an Iranian 33-year-old female patient.

2. Case Presentation

A 33-year female Iranian patient referred to the emergency department of Gastroenterology and liver diseases Research Center, Shahid Beheshti University of Medical Sciences with extreme abdominal pain, nausea and constipation. The patient did not have a history of IBD, FAP or HNPCC. However, she had been referred to the Gastroenterology and Liver Diseases Research Center, Shahid Beheshti University of Medical Sciences, Tehran, Iran due to abdominal pain three years ago and multiple polyps in stomach were detected. Plain abdominal CT

Copyright @ 2015, Colorectal Research Center and Health Policy Research Center of Shiraz University of Medical Sciences. This is an open-access article distributed under the terms of the Creative Commons Attribution-NonCommercial 4.0 International License (http://creativecommons.org/licenses/by-nc/4.0/) which permits copy and redistribute the material just in noncommercial usages, provided the original work is properly cited.

scan revealed a mass suggesting intussusception in the jejunum. Axial spiral Modified Card Sorting Test (MCST) of thorax, abdomen and pelvis with intravenous and oral contrast had normal findings. Balloon association enteroscopy showed two 10 mm and 40 mm polyps in jejunum. After injection, larger polyps were excised and retrieved. The pathological diagnosis of polyp was villous adenoma with mild grade dysplasia (Figure 1). Tumor invaded to muscularis propria PT2. Immunohistochemistry (IHC) had positive results for chromogranin, patchy for Epithelial Membrane Antigen (EMA), Carcinoembryonic Antigen (CEA) and 2-3% Ki67 and negative for neuron-specific enolase (NSE) and synaptophysin in tumor cells. Exploratory laparotomy revealed jejunojejunal intussusceptions and exploratory laparotomy with jejunoileal anastomosis was performed. Further follow-up of patients had good clinical results.

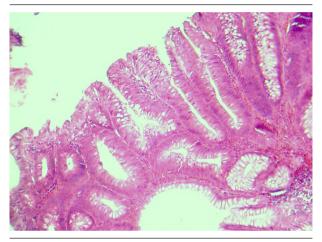


Figure 1. Pathological Diagnosis of Jejunal Polyp With Villous Adenoma Feature in an Adult Jejunojejunal Intussusception Case

3. Discussion

Intussusception is characterized by navigation of a part of gastrointestinal tract into an adjacent section (6). Intussusception is so rare in adults and small intestine intussusception in adults is usually due to benign entity; whereas, in large intestine malignant neoplasms should be considered (3). Jejunojejunal intussusception in adults is a rare entity and the incidence of intussusception in jejunum is higher than ileum. In this case report, we presented a rare case of jejunojejunal intussusception caused by a jejunal villous adenoma polyp in a 33year old Iranian female. Of all small intestinal adenoma lesions, up to 20% occur in the small intestine (7). Among adenoma polyps, villous adenoma with its invasive histological features has the highest risk of progression to cancer than other adenomas (8). In line with our study, Takeuchi et al. reported a rare case of a 73-year-old male with tubulovillous adenocarcinoma presenting with intussusception. The patient complained with vomiting, nausea and abdominal distension. Plain abdominal CT and further evaluation revealed intussusception in the jejunum. Surgery was performed and a jejunal tumor of about 30 cm from the treitz ligament was detected (9). In another study, Neishaboori et al. reported a 40-year-old Iranian female with Jejunal intussusception caused by a huge inflammatory fibroid polyp (Vanek's tumor). The patient presented with severe, postprandial abdominal pain followed by projectile vomiting over three days. In consistent to our study, they performed laparotomy and observed an 18 cm tumor from the distal jejunum causing jejunojejunal intussusceptions. The intussuscepted segment was resected and end-to-end anastomosis was performed (10). Basu et al. reported a case with jejunojejunal intussusception raised by a Jejunal Gastrointestinal Stromal Tumor (GIST). In their study, a 46-year-old female presented with abdominal pain, anorexia, mild abdominal distension, constipation and vomiting. As a surgical treatment they performed formal midline laparotomy and found a 10-cm long jejunojejunal intussusception 60 cm distal to the duodenojejunal junction without perilesional adenopathy (11). There is still a diagnosis challenge in Intussusception cases since they usually have nonspecific symptoms and no specific clinical presentations. These unspecific symptoms often lead to a late diagnosis. Imaging technics including ultrasonography and CT are considered as best modalities (12, 13). Even with the help of these advanced imaging technics, most cases of intussusception are diagnosed on the surgical operation procedures. The treatment of intussusception in adult cases is surgical operation. Recently, laparoscopic procedure has been demonstrated as a minimally invasive technique in adult cases. As in this case, we performed exploratory laparotomy with jejunoileal anastomosis. Martis et al. reported a rare case of jejunojejunal intussusception raised by GIST in a 50-year-old female presenting with abdominal pain, nausea and vomiting (14). In the present case report, the patient referred with extreme abdominal pain and diarrhea. This is similar to Martis et al. report (14). However, in most cases these unspecific presentations and symptoms usually result in a late diagnosis. In another case report, Choi et al. reported a 36-year-old female with Peutz-Jeghers syndrome (PJS) with a large jejunal polyp presenting as an asymptomatic small bowel intussusception identified on CT. In that case, they performed Double Balloon Enteroscopy (DBE) to remove the lesions. In contrast to our study, pathologic feature of polyps they detected was hamartomatous (15). In a case repot, Singhal reported a 51-year-old adult with jejunoileal intussusception caused by an Inflammatory Fibroid Polyp (IFP). In double contrast CT scan, they found a mass of jejunoileal intussusception. Consistent to our study, they performed laparotomy and observed a jejunoileal intussusception of about 14 cm length involving the distal jejunum and proximal ileum (16). In conclusion, adult intussusception caused by villous adenoma is extremely rare. Since it has nonspecific symptoms, this might result in a late diagnosis. Therefore, imaging modalities including CT scan play a significant role in diagnosis. Surgical operation is the best treatment in adult intussusception with best results.

Authors' Contributions

Seyed Mohammad Hossein Kashfi: writing the manuscript;Faegheh Behboudi Farahbakhsh: sectioning the specimen and collecting the data; Mina Golmohammadi : sectioning the specimen and collecting the data; Ehsan Nazemalhosseini Mojarad: Revising the manuscript; Pedram Azimzadeh: Revising the manuscript; Shahrokh Iravani: follow-up of patient; Katayoun Gohari Moghadam: Pathology diagnosis of the specimen; Mohammad Reza Zali: Diagnosis of intussusception in this case; Hamid Asadzadeh Aghdaei: gave the idea of reporting this case report.

Funding/Support

This study was supported by the Gastroenterology and Liver Diseases Research Center, Shahid Beheshti University of Medical Sciences, Tehran, Iran.

References

- Wang N, Cui XY, Liu Y, Long J, Xu YH, Guo RX, et al. Adult intussusception: a retrospective review of 41 cases. World J Gastroenterol. 2009;15(26):3303-8.
- Azar T, Berger DL. Adult Intussusception. Ann Surg. 1997;226(2):134– 8.
- Eisen LK, Cunningham JD, Aufses AJ. Intussusception in adults: institutional review. J Am Coll Surg. 1999;188(4):390–5.
- 4. Gayer G, Zissin R, Apter S, Papa M, Hertz M. Pictorial review: adult

intussusception-a CT diagnosis. Br J Radiol. 2002;**75**(890):185-90.

- Yamada H, Morita T, Fujita M, Miyasaka Y, Senmaru N, Oshikiri T. Adult intussusception due to enteric neoplasms. *Dig Dis Sci.* 2007;52(3):764–6.
- Gayer G, Apter S, Hofmann C, Nass S, Amitai M, Zissin R, et al. Intussusception in adults: CT diagnosis. *Clin Radiol*. 1998;53(1):53–7.
- Perzin KH, Bridge MF. Adenomas of the small intestine: a clinicopathologic review of 51 cases and a study of their relationship to carcinoma. *Cancer*. 1981;48(3):799–819.
- Winawer SJ, Zauber AG, Ho MN, O'Brien MJ, Gottlieb LS, Sternberg SS, et al. Prevention of colorectal cancer by colonoscopic polypectomy. The National Polyp Study Workgroup. N Engl J Med. 1993;329(27):1977-81.
- Takeuchi N, Semba S, Naba K, Aoki R, Nishida Y, Nomura Y, et al. Jejunal tubulovillous adenocarcinoma in adenoma presenting with entero-enteric intussusception. *Clin J Gastroenterol.* 2013;6(1):46–9.
- Neishaboori H, Maleki I, Emadian O. Jejunal intussusception caused by a huge Vanek's tumor: a case report. *Gastroenterol Hepa*tol Bed Bench. 2013;6(4):210–3.
- Basu A, Dutta M, De U, Biswas S. Jejunojejunal intussusception caused by a jejunal gastrointestinal stromal tumour (GIST). *Hell J* Surg. 2014;86(1):37-41.
- Urbano J, Serantes A, Hernandez L, Turegano F. Lipoma-induced jejunojejunal intussusception: US and CT diagnosis. *Abdom Imaging*. 1996;21(6):522-4.
- 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(3):733-44.
- Martis JJ, Rajeshwara KV, Murulya KS, Raghavendra BK, Alex KM. A rare cause of jejunojejunal intussusception in an adult. *Indian J* Surg. 2013;75(Suppl 1):18–20.
- Choi SJ, Shin SJ, Choi JW, Song JC, Lim SG, Lee KM, et al. Small bowel intussusception caused by a jejunal polyp in Peutz-Jeghers syndrome: a case treated with double balloon enteroscopy. *Dig Dis Sci.* 2011;56(3):915–8.
- Singhal BM, .Kumar V, Sagar S, Singh CP. Adult jejunoileal intussusception caused by inflammatory fibroid polyp. *Niger J Surg Sci.* 2013;23(1):26.