Adult intussusceptions caused by a lipoma in the jejunum: report of a case and review of the literature

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Abstract
Intussusceptions in adults is rare. Gastrointestinal lipomas are rare benign tumors and intussusceptions due to a gastrointestinal lipoma constitutes an infrequent clinical entity. Lipoma may develop as a benign tumor in all organs and rarely in large or small intestine. The present report describes a case of jejunojejunal intussusceptions in an adult with a history of colicky upper abdominal pain. Ileo-ileal invagination was diagnosed by computed tomography scan. Exploratory laparotomy revealed jejunojejunal intussusceptions secondary to a lipoma which was successfully treated with segmental intestinal resection. A review of the literature is also performed regarding this rare association revealing the diagnostic and therapeutic debates that exist.

Abstract (french)
L’invagination chez les adultes est rare. Les lipomes gastro-intestinaux sont de rares tumeurs bénignes et l’invagination intestinale due à un lipome gastro-intestinal constitue une entité clinique trés rare. Le lipome peut se développer comme une tumeur bénigne dans tous les organes et rarement dans l’intestin grêle ou le colon. Le présent rapport décrit un cas d’invagination jéjunöjéjunale chez un adulte avec une histoire de douleurs abdominales. Iléo-iléale invagination a été diagnostiquée par tomodensitométrie. Une laparotomie exploratrice a révélé l’existence d’une invagination jéjunöjéjunale secondaire à un lipome qui a été traitée avec succès par une résection intestinale segmentaire. Une revue de la littérature est également effectuée au sujet de cette association rare révélant les débats diagnostiques et thérapeutiques qui existent.

Keywords: Intussusceptions, Jéjunal lipoma, Intestinal tumor, Surgery

Background
Intussusceptions was reported for the first time in 1674 by Barbette of Amsterdam [1]. The occurrence of intussusceptions in adults is rare, accounting for less than 5% of all cases of intussusceptions and almost 1%-5% of bowel obstruction [2]. In contrast to pediatric intussusceptions, which is idiopathic in 90% of cases, adult intussusceptions has an organic lesion in 70% to 90% of cases [3]. The majority of lipomas in the small bowel are solitary. Approximately 5% are multiple [4]. Symptomatic lipoma manifestations are hemorrhage or intestinal obstruction. Due to their intramural location, lipomas can also serve as the leading point for intussusceptions. We report a rare case of jejuno-jejunal intussusceptions in an adult secondary to an jejunal lipoma.

Case presentation
A 35-year-old man was admitted to the emergency department in a tertiary referral hospital with 4 months history of intermittent upper abdominal pain accompanied with nausea. The patient had no past history of peptic ulcer disease, alteration in bowel habits, melena or
weight loss. On examination, he was apyrexial and hemodynamically stable. His abdomen was distended and no palpable abdominal masses; bowel sounds were hyper audible. Initial A rectal examination revealed no masses or blood. Laboratory blood tests were normal. Abdominal radiography revealed prominent dilatation of the small bowel with air fluid levels (Figure 1). Abdominal CT showed a target sign- or sausage-shaped lesion typical of an intussusceptions that varied in appearance relative to the slice axis (Figure 2). The inner central area represented the invigilated intussuscepted, surrounded by its mesenteric fat and associated vasculature, and all surrounded by the thick-walled intussuscipliens. More head-side scans showed a low-density homogenous mass measuring 4 cm that was considered to be the leading point for the invagination (Figure 3). These findings led to a diagnosis of intussusceptions induced by a tumor most likely begin. The decision was made to undertake an urgent exploratory laparotomy. At laparotomy, 50 cm distal to the ligament of Treitz, a jejuno-jejunal intussusceptions was identified. We conducted a desinvagination Benin saw the character of the lesion on CT. The presence of irreversible ischemia in a small portion of the intussusceptum necessitated segmental resection and primary anastomosis (Figure 4). The postoperative period was uneventful and the patient was discharged on the sixth postoperative day. Gross examination of the respected specimen revealed a round tumor covered with mucosa measuring 6 cm. A microscopic examination revealed fat cells proliferating in the submucosal layer and confirmed the diagnosis of ileal lipoma (Figure 5).

The histopathology report confirmed a 60-mm submucosal lipoma in the jejunum as a cause for a 30-cm jejuno-jejunal intussusceptions. There was no evidence of dysplasia or malignancy.

Discussion

Intussusceptions in adulthood is unusual, with an incidence of approximately 2-3 cases per population of
1,000,000 per year [5]. The most common classification system divides intussusceptions into four categories: enteric, ileocolic, ileoceleal and colonic [1-4]. In adults, intussusceptions is more likely to present insidiously with vague abdominal symptoms and rarely presents with the classic triad of vomiting, abdominal pain and passage of blood per rectum, making diagnosis difficult [6]. Tumors of the small bowel account for only 1% to 2% of all gastrointestinal tumors, and benign tumors account for approximately 30% of all small-bowel tumors [7]. Lipomas are benign tumors of mesenchymal origin. They are the second most common benign tumors in the small intestine and account for 10% of all benign gastrointestinal tumors and 5% of all gastrointestinal tumors [1,2,5]. Gastrointestinal lipomas are most commonly located in the colon (65% to 75%), small bowel (20% to 25%) and occasionally in the foregut (< 5%) [8]. Fifty-one cases of adult intussusceptions induced by a lipoma, including our present case, have been reported in the English literature during the past decade (Table 1) [9]. Lipomas are largely asymptomatic. The majority of presenting features are either intestinal obstruction or hemorrhage [1,2,5-8]. Their usual location in the small intestine is ileum (50%) while jejunum is the least common. The peak age of incidence is in the 6th-7th decades of life and it appears that females are more prone to lipomas. Malignant degeneration has never been reported [5]. The clinical presentation is very nonspecific which makes this a difficult condition to diagnose. According to the literature, only 32% to 50% of cases are diagnosed preoperatively, despite the evolution of imaging methods [9-11]. Abdominal pain, nausea, diarrhea and bleeding per rectum are the common symptoms. Rarely, this can present with acute intestinal obstruction. The classical triad of abdominal pain, sausage shaped palpable mass and passage of red current jelly stools seen in children is rarely seen in adults. Fewer than 20% of cases present acutely with complete bowel obstruction. A palpable abdominal mass is present in only 7% to 42% of cases [12,13]. Lipomas can be diagnosed through conventional endoscopy, capsule endoscopy, barium studies and, most importantly, CT scan [14]. Ultrasound is usually the first modality to be recruited. However, it is operator-dependent and the presence of distended bowel decreases the ability to demonstrate the site of the obstruction. Computed tomography is the imaging method of choice for diagnosing intussusceptions. A submucosal lipoma can be diagnosed if a smooth well-circumscribed mass of fat density (-50 to -100 Hounsfield Units) is revealed within the lumen of the bowel or intussuscipiens. The sensitivity of CT scan to correctly diagnose intussusceptions has been reported from 71.4%-87.5% while its specificity in adults has been reported to be 100% as verified by the subsequent surgery [14,15]. Capsule endoscopy and digital balloon endoscopy are newer means for diagnosing lipomas and are particularly helpful in cases involving small bowel lipomas [8]. Definitive surgical resection remains the recommended treatment for adult intussusceptions due to the large proportion of structural causes and the relatively high incidence of malignancy; however, the optimal surgical management remains controversial [1,2,6,7,9]. Some investigators have stated that small bowel intussusceptions should still be reduced only in patients in whom a definitive benign diagnosis has been made preoperatively, or in patients in whom resection may result in short gut syndrome [9]. The dangers of transperitoneal, vascular, and intraluminal seeding after exposing and handling friable and edematous malignant tissues has led many surgeons to
Table 1 The characteristics of the reported cases of adult intussusception induced by a lipoma

| Case | Age  | Gender | Diagnostic modality | Tumor location | Size (cm) | Reference                          |
|------|------|--------|---------------------|----------------|-----------|-----------------------------------|
| 1    | 69   | Male   | US, CS              | Descending colon | 4         | J Clin Ultrasound                  |
| 2    | 42   | Male   | CS, BE, CT          | Descending colon | 4.5       | Am Surg                           |
| 3    | 39   | Male   | US, CT              | Ileum           | 4         | J Korean Med Sci                   |
| 4    | 72   | Male   | EGD, US, CT         | Stomach         | 10        | Dig Surg                          |
| 5    | 28   | Male   | CT                  | Jejunum         | 3         | Ann R Coll Surg Engl              |
| 6    | 20   | Female | CT                  | Ileum           | 18        | Emerg Radiol                      |
| 7    | 41   | Male   | CT                  | Ileum           | ND        | Australas Radiol                  |
| 8    | 44   | Female | CT, CS, ECS         | Ileum           | 5         | Abdom Imaging                     |
| 9    | 51   | Female | US, ECS, CT         | Cecum           | 10        | Rom J Gastroenterol               |
| 10   | 56   | Male   | US, CT              | Ascending colon | 6         | J Laparoendosc Adv Surg Tech A    |
| 11   | 50   | Male   | ECS, CS, CT         | Ascending colon | 5         | Pathol Int                        |
| 12   | 72   | Male   | CT, EGD             | Stomach         | 6         | Can J Gastroenterol               |
| 13   | 55   | Male   | CT                  | Ileum           | ND        | Surg Today                        |
| 14   | 63   | Female | US, CT              | Ileum           | 2.5       | Surg Today                        |
| 15   | 73   | Female | ECS, MRI            | Sigmoid colon   | ND        | Arch Surg                         |
| 16   | 63   | Male   | CT                  | Ileum           | 3         | JSLS                              |
| 17   | 85   | Male   | US, CT              | Jejunum         | 4         | J Gastroenterol Hepatol           |
| 18   | 62   | Male   | CT, CS              | Sigmoid colon   | 3.5       | Dig Dis Sci                      |
| 19   | 55   | Female | CT                  | Transverse colon | 12        | Am Surg                           |
| 20   | 31   | Female | CT                  | Ascending colon | 5         | Can J Surg                        |
| 21   | 47   | Female | US, CT              | Ileum           | 5         | Ullus Travma Aci Ileum Derg       |
| 22   | 56   | Female | US, CS, CT          | Transverse colon | 5         | Ullus Travma Aci Ileum Derg       |
| 23   | 64   | Male   | CS, CT              | Transverse colon | 6         | Clin Gastroenterol Hepatol        |
| 24   | 55   | Male   | CT, ECS             | Jejunum         | 4         | World J Gastroenterol             |
| 25   | 42   | Male   | US, CT              | Ileum           | 3         | Case Rep Gastroenterol            |
| 26   | 47   | Female | CT                  | Ileum           | 3         | J Laparoendosc Adv Surg Tech A    |
| 27   | 47   | Female | CT, CS, Enema       | Ascending colon | 5         | Endoscopy                         |
| 28   | 36   | Male   | CS, CT, ECS         | Ileum           | 9         | Cases J                           |
| 29   | 36   | Male   | CT, ECS             | Ileum           | 4         | J Nippon Med Sch                  |
| 30   | 82   | Male   | CS, CT              | Sigmoid colon   | 8         | Gastrointest Endosc               |
| 31   | 69   | Male   | CT, CS              | Transverse colon | 7         | Dig Dis Sci                      |
| 32   | 38   | Female | CS, CT              | Ileum           | 3.3       | Clin Gastroenterol Hepatol        |
| 33   | 38   | Female | US, CT, CS          | Cecum           | 6         | Emerg Radiol                      |
| 34   | 45   | Male   | CT                  | Ileum           | 2.5       | N Engl J Med                      |
| 35   | 43   | Female | CS, CT              | Ascending colon | 5         | Rev Esp Enferrm Dig               |
| 36   | 57   | Female | CS, CT              | Transverse colon | 5.5       | Rev Esp Enferrm Dig               |
| 37   | 51   | Male   | US, CT, CS          | Ileum           | 3         | Gastroenterology                  |
| 38   | 77   | Male   | CT                  | Cecum           | 3.5       | JLS                              |
| 39   | 46   | Male   | CS, CT, ECS         | Descending colon | 6         | Endoscopy                         |
| 40   | 33   | Male   | CT, CS, BE          | Ileum           | 4         | Case Rep Gastroenterol            |
| 41   | 32   | Female | CT                  | Ascending colon | 5.8       | Gastroenterology                  |
| 42   | 49   | Male   | US, CT              | Descending colon | 5         | Gastroenterology                  |
| 43   | 53   | Female | US, CS, ECS         | Ascending colon | 7         | Medicina (Kaunas)                |
| 44   | 26   | Female | CT                  | Ileum           | ND        | Am J Surg                         |
| 45   | 51   | Female | CT                  | Transverse colon | 6.2       | J Gastroenterol Hepatol           |
| 46   | 68   | Male   | CS                  | Jejunum         | 3.2       | World J Gastroenterol             |
advocate en bloc resection of the lesion. All surgeons agree, though, that reduction should not be attempted if there are signs of irreversible bowel ischemia, inflammation or when malignancy is being suspected [5,16,17]. The advantages of intraoperative reduction of the intussusceptions prior to resection, especially when the small bowel is affected, are that it may preserve a considerable length of bowel and thereby prevent development of short-bowel syndrome.

**Conclusion**

The lipoma is a rare benign tumor of the digestive tract. The diagnosis of intussusceptions in adults can be difficult because of atypical and episodic symptoms. A high level of clinical suspicion and an abdominal CT scan are most useful tools for making a timely diagnosis. Surgical resection remains the treatment of choice and produces an excellent prognosis.

**Consent**

Written informed consent was obtained from the patient for publication of this case report and accompanying images.

**Abbreviations**

CT: Computed tomography; MR: Magnetic resonance imaging; CS: Colonoscopy; ECS: Enema contrast study; EGD: Esophagogastroduodenoscopy; US: Ultrasonography; ND: Not described.

**Competing interests**

The authors declare that they have no competing interests.

**Authors’ contributions**

All of the authors were involved in the preparation of this manuscript. OM performed the operation and revised the manuscript. HH was an assistant surgeon and made substantial contributions to conception and design. LC described histological finding and was involved in drafting the manuscript. All authors read and approved the final manuscript.

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