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Adult intussusception

An overlooked diagnosis in the emergency department

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ABSTRACT

الأهداف: دراسة وتحليل المسببات والأعراض السريرية وكذلك طرق التشخيص وخيارات العلاج المختلفة لهذا المرض النادر الحدوث.

الطريقة: تمت هذه الدراسة الإستعادية على 14 حالة أدخلت مستشفى سوهاج الجامعي بمصر وكذلك مستشفى القوات المسلحة بنجران بالمملكة العربية السعودية خلال الفترة من 2004م حتى 2012م وقد تم تشخيصها إما قبل أو أثناء العملية على أنها حالات تداخل الامعاء الانغلاقى في الكبار.

النتائج: تراوحت أعمار المرضى ما بين 22–63 عاما بمتوسط46 عام وكانت نسبة الذكور منهم 71.4%. كانت الشكوى الرئيسية في92.9% من المرضى هي آلام متكررة بالبطن. تم تشخيص جميع الحالات قبل العملية بواسطة الاشعة المقطعية وتأكد التشخيص أثناء العملية. تم التعرف على الأسباب في جميع الحالات ماعدا حالتين. تركزت معظم الحالات في الامعاء الدقيقة بنسبة (64.3%) وكان السبب الرئيسي لهذه الحالات هي أورام حميدة بينما باقي الحالات كانت بالقولون سجلت حالتين منها أورام سرطانية. كانت الجراحة هي الحل الامثل في جميع الحالات وكانت نسبة المضاعفات بعد العملية 3 (21.4%) وهناك حالة وفاة كانت تعاني من ورم سرطاني بالقولون حدث لها تسرب لمحتويات الأمعاء بالبطن وتعفن في الدم مع فشل في أجهزة الجسم الحيوية.

خاتمة: أن تشخيص هذا المرض يحتاج إلى درجة عالية من الشك من قبل طبيب الرعاية الاولية وطبيب الطوارئ وكذلك الجراحين وخاصة للمرضى الذين يترددون على قسم الطوارئ بآلام متكررة بالبطن. ثبت ايضا من البحث أن الاشعة المقطعية تلعب دوراً أساسياً في التشخيص كما أن العلاج الجراحي هو الطريقة المثلى في التعامل مع مثل هذه الحالات.

Objectives: To study the appropriate method of diagnosis and management in adult intussusception (AI) focusing on the clinical manifestations, diagnostic tools, and management of this rare disease.

Methods: This retrospective study reviewed and analyzed the demographic data, clinical features, diagnosis, management, and pathology reports of all adult patients (18 years of age and older) with a diagnosis of intussusception admitted to Sohag University Hospital, Sohag, Egypt, and Najran Armed Forces Hospital, Najran, Kingdom of Saudi Arabia (KSA) from January 2004 to August 2012.

Results: From 2004-2012, 14 patients with AI were diagnosed and treated. Ages ranged from 22-63 years. Ten patients (71.4%) were males. Thirteen patients (92.9%) presented with abdominal pain. All patients were diagnosed after a CT scan, and confirmed at laparotomy. The lead point was found in all, except for 2 patients. Ileo-ileal and jejuno-jejunal intussusceptions represented most of our cases (64.3%), followed by ileocolic (28.5%), and colocolic (7.1%). The most common causes were Peutz-Jeghers polyps (5 cases), submucosal lipoma (2), and malignancy (2). Surgery was the treatment option in all except one patient. Postoperative complications occurred in 3 cases (21.4%). One patient died 28 days postoperatively due to septic shock and multiorgan failure.

Conclusion: Owing to its rarity, AI needs a high index of suspicion especially in patients attending ERs with recurrent abdominal pain. A CT scan is of prime importance in the diagnosis, and surgical treatment is the preferred method of management.

Intussusception is defined as the telescoping of a segment of the gastrointestinal tract into an adjacent one.[1] Intussusception in adults is rare, accounting

for only 5% of all intussusceptions, 1% of all bowel obstruction, and 0.003-0.02% of all hospital admissions.[2] Neoplasm is the most common cause, and is found in approximately 65% of adult cases.[3] Patients most commonly present with abdominal pain, nausea, and vomiting. Less frequently, melaena, weight loss, fever, constipation, or occasionally acute abdomen is the presentation.[4] Intussusception is usually diagnosed with CT, which shows a pathognomonic bowel-within-bowel configuration, with, or without contained fat and mesenteric vessels.[3] According to the literature, most patients with adult intussusceptions (AI) require surgical resection as most has a definable pathology. However, there is no consensus regarding the extent of resection and reduction before excision. Controversy regarding the appropriate surgical management of AI, as well as its rarity is the rationale behind conducting our study to highlight the disease, and study the appropriate method of its diagnosis and management. We reviewed cases of AI focusing on the clinical manifestations, diagnostic tools, and management of this rare disease that can easily be missed, especially in patients attending the emergency department (ER).

Methods. This retrospective study reviewed and analyzed the demographic data, clinical features, diagnosis, management, and pathology reports of all adult patients (18 years of age and older) with a diagnosis of intussusception admitted to Sohag University Hospital, Sohag, Egypt, and Najran Armed Forces Hospital, Najran, Kingdom of Saudi Arabia (KSA) from January 2004 to August 2012. Ethical approval was obtained for the local ethics committee prior to the commencement of the study, and informed patient consent was received from all study participants. An intussusception that involved only the jejunum or ileum was considered an enteric intussusception. An intussusception that involved the ileum and colon was designated as an ileocolic intussusception. An intussusception that involved only the colon was considered a colocolic intussusception, and one that involved the sigmoid colon and rectum was considered a sigmoidorectal intussusception.[5] Acute symptoms were defined as <4 days, subacute symptoms were defined as 4-14 days, and chronic symptoms were defined as >14 days.[6] Diagnosis was based on clinical suspicion and imaging investigations including abdominal plain x-rays, abdominal ultrasonography (US), and CT. Colonoscopy was performed in selected patients who presented with bleeding per rectum, or signs of large bowel obstruction. The following were the inclusion

criteria: patients presenting with hematochezia (passage of maroon or bright red blood, or blood clots per rectum), and, or large bowel obstruction noted clinically, and on abdominal plain films. The exclusion criteria were: suspected upper gastrointestinal source of bleeding, for example, history of hematemesis/malena, or gastric aspirates containing coffee-ground material, or bright red blood; hemorrhoidal bleeding established by per rectum exam/proctoscopy; and rectal bleeding as a result of acute infectious bloody diarrhea. The final diagnosis was made intraoperatively after exploration, and defining the site, and cause of intussusception. Histopathology confirmed the nature of the lesion, whether benign, or malignant. Under general endotracheal anesthesia, all patients were treated surgically by midline abdominal incision, resection of involved segments, and primary anastomosis. Postoperative morbidity and mortality were analyzed.

Results. A total of 14 adult patients were identified with a diagnosis of intussusception. The average patients' age was 46.9 years (range: 22-63 years). Ten patients (71.4%) were males, and 4 (28.6%) were females. Eleven (78.6%) patients had chronic symptoms, and 3 (21.4%) had acute presentation (large bowel obstruction [one patient]), severe rectal bleeding (one patient), and small bowel ischemia secondary to portal, splenic, and superior mesenteric vein thrombosis (one patient). Abdominal pain was the most common complaint in 13 (92.9%) patients. Other symptoms encountered were nausea, vomiting, constipation, and rectal bleeding. Abdominal examination was unremarkable in chronic cases with no palpable masses, while there was abdominal distension and tenderness in patients presenting with signs of bowel obstruction. The mean duration of symptoms was 9.6 (range: one to 18) weeks. Patients' characteristics, operative findings, morbidity, and mortality are shown in **Table 1**. A plain x-ray of the abdomen was carried out in the ER as an initial investigation. It was inconclusive in 12 patients, and suggestive of intestinal obstruction in 2 patients. Abdominal CT scan (plain, and with contrast) was performed in all patients. It confirmed the diagnosis of intussusception in all of them, and the suggestive findings revealed inhomogeneous soft tissue mass, target, or sausage shaped. The causative pathology, such as polyps (**Figure 1**), lipoma (**Figures 2A & 2B**), or tumoral lesions were identified in 12 patients. In one patient, ileocolic intussusception was found, in addition to portal, splenic, and superior mesenteric veins thrombosis causing small bowel ischemia. The causative lesion was not established in this patient.

Table 1 - Characteristics of 14 cases of adult intussusceptions included in a study at Sohag University Hospital, Sohag, Egypt, and Najran Armed Forces Hospital, Najran, Kingdom of Saudi Arabia.

| No.	Age (years)	Gender	Presentation	Location	Treatment	Pathology	Postoperative complications	Mortality
1	22	Male	Rectal bleeding, abdominal pain	Ileo-ileal	Resection anastomosis	Peutz-Jeghers polyps	---	No
2	50	Female	Abdominal pain	Jejuno-jejunal	Resection anastomosis	Gastrointestinal stromal tumor	---	No
3	42	Male	Abdominal pain	Ileo-ileal	Resection anastomosis	Submucosal lipoma	---	
4	33	Male	Abdominal pain	Ileocolic	Right hemicolectomy	Idiopathic	Wound sepsis, mild leakage	No
5	55	Female	Abdominal pain	Jejuno-jejunal	Resection anastomosis	Peutz-Jeghers polyps	---	No
6	60	Female	Bowel obstruction, constipation	Colocolic	Left hemicolectomy + primary anastomosis	Adenocarcinoma left colon	Leakage, sepsis, septic shock, multiple organ failure	Yes
7	45	Male	Abdominal pain	Jejuno-jejunal	Resection anastomosis	Peutz-Jeghers polyp	---	No
8	50	Male	Abdominal pain	Ileo-ileal	Resection anastomosis	Inflammatory fibroid polyp	---	No
9	52	Male	Abdominal pain	Ileo-ileal	Resection anastomosis	Meckel's diverticulum	---	No
10	48	Male	Abdominal pain	Ileo-ileal	Resection anastomosis	Peutz-Jeghers polyps	---	No
11	58	Female	Abdominal pain, diarrhea	Ileocolic	Right hemicolectomy	Submucosal lipoma	Bleeding, re-exploration, wound sepsis	No
12	38	Male	Abdominal pain	Ileo-ileal	Resection anastomosis	Peutz-Jeghers polyps	---	No
13	40	Male	Acute recurrent, abdominal pain	Ileocolic + jejunal bowel ischemia	Resection anastomosis of ischemic segment	Idiopathic	---	No
14	63	Male	Abdominal pain, diarrhea	Ileocolic	Right hemicolectomy	Primary adenocarcinoma Cecum	---	No
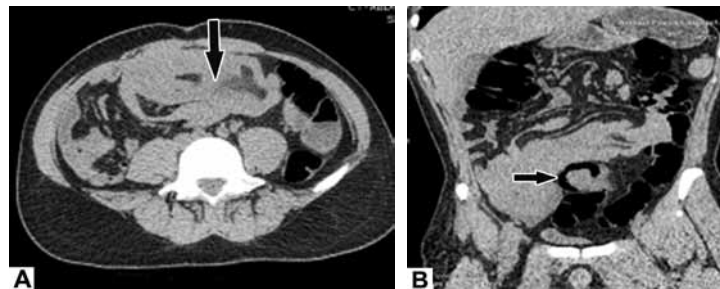
Figure 1 - An image showing: **A)** axial plain CT scan of the abdomen showing jejuno-jejunal intussusception with pseudokidney appearance (arrow), and **B)** coronal reconstruction demonstrating the intussusception, as well as the sigmoid colon polyp (arrow).
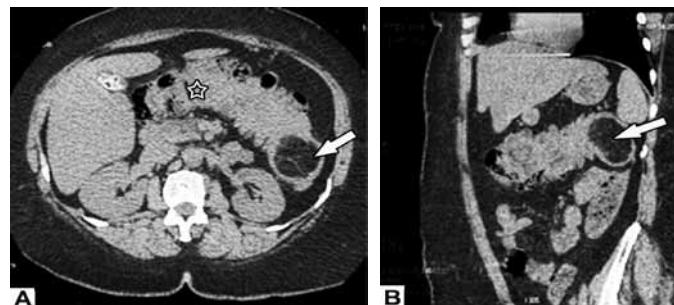

Figure 2 - Scan images of the abdomen demonstrating the ileocolic intussusception (star) with the head of the intussusceptum containing fatty densities (lipoma)(arrow): **A)** plain axial, and **B)** sagittal CT.
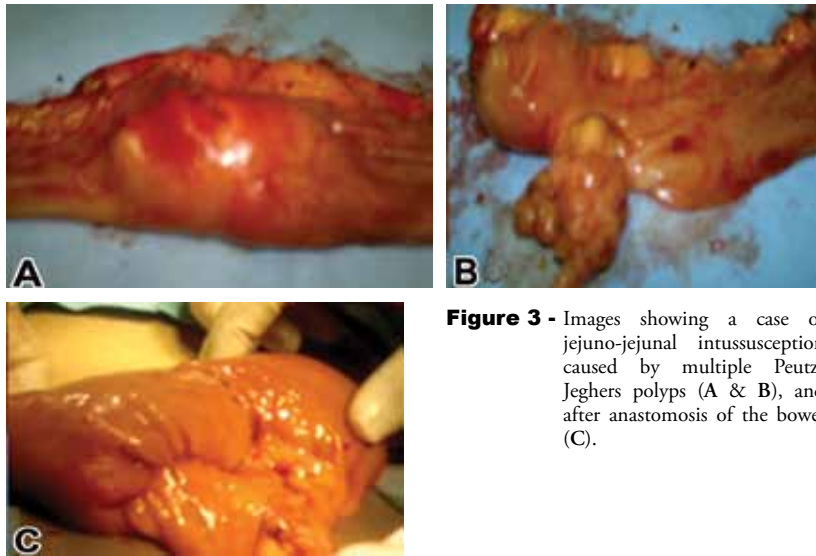
Figure 3 - Images showing a case of jejuno-jejunal intussusception caused by multiple Peutz-Jeghers polyps (**A** & **B**), and after anastomosis of the bowel (**C**).
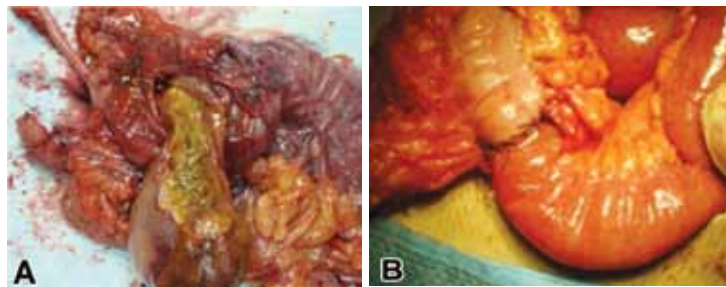

Figure 4 - Images showing a case of ileocolic intussusception caused by a submucosal lipoma (ulcerated) (**A**) and ileotransverse anastomosis after a right hemicolectomy (**B**).
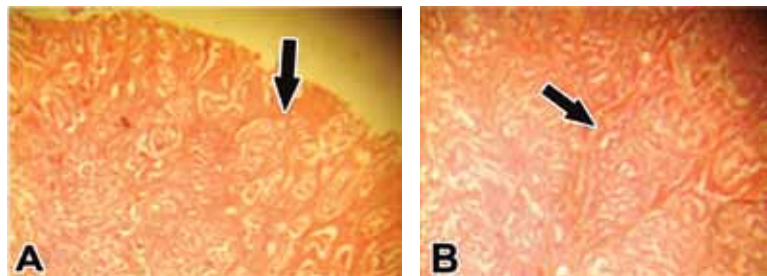

Figure 5 - Images showing hamartomatous polyps in a case of Peutz-Jeghers syndrome causing intussusception of the small gut. A large polyp projecting into the lumen (Hematoxylin and Eosin low power view); **A**) proliferation of numerous benign glands lined by typical simple columnar epithelium (arrow), and **B**) pseudo-invasion of glands in proliferating fibromuscular stroma (arrow).
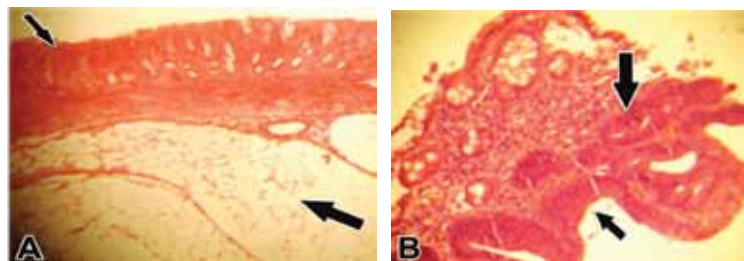

Figure 6 - Images showing a case of intussusception presenting with: **A**)large submucosal lipoma (big arrow) beneath the ulcerated mucosa (small arrow); and **B**) adenocarcinoma showing invasion of lamina propria (big arrow), at its junction with normal mucosa (small arrow).

Colonoscopy was performed in 3 patients and revealed a sigmoid polyp in one, carcinoma of the left colon in another, and normal findings in the third. Ileo-ileal and jejuno-jejunal intussusceptions represented most of our cases (64.3%), followed by ileocolic (28.5%), and colocolic (7.1%). Ileocolic intussusception was diagnosed in 4 patients, and colocolic in one patient. All patients underwent laparotomy exploration through a midline incision, and were managed according to the findings. In 9 patients with small bowel intussusception, the treatment was reduction of intussusception, and limited resection of the diseased segment of the bowel (**Figures 3A, 3B, & 3C**). In 4 patients with ileocolic intussusceptions, right hemicolectomy was performed in 3 patients (**Figures 4A & 4B**), while in the fourth patient, intussusception was reduced spontaneously during the CT contrast study. In this case, the CT also revealed small bowel ischemia involving mostly the jejunum secondary to portal, splenic, and superior mesenteric vein thrombosis. Resection of the ischemic bowel was performed, and the patient was maintained on intravenous heparin infusion. One patient with adenocarcinoma of the left colon underwent primary resection (left hemicolectomy) without colostomy. Urgent colonoscopy after resuscitation was performed in one patient that presented with severe rectal bleeding. It revealed a large polyp in the sigmoid colon, however, it was not the source of bleeding. The abdominal CT scan revealed jejuno-jejunal intussusception with multiple small and large intestinal polyps. This patient underwent surgical exploration, and resection of the involved small bowel. Six months later, colonoscopic polypectomy of the sigmoid polyp without the need for further surgical intervention was performed. Pathological causes were identified in 12 (85.7%) patients. Benign lesions were detected in all enteric, and 2 ileocolic intussusceptions as follows: Peutz-Jeghers polyps (5 cases) (**Figures 5A & B**), submucosal lipoma (2 cases) (**Figure 6A**). Other causes include gastrointestinal stromal tumor (GIST), Meckel's diverticulum, and inflammatory fibroid polyp (one case each). Malignancy in the form of primary adenocarcinoma was found in 2 cases: ileocecal intussusception with cecal cancer, and colocolic intussusception on top of left colon cancer (**Figure 6B**). Postoperative complications occurred in 3 cases (21.4%). One patient had mild wound sepsis and leakage, which improved with conservative measures. Another patient developed bleeding per rectum, 12 days following right hemicolectomy. This patient was readmitted, explored, and the bleeding site at the anastomotic line was sutured and the bleeding

stopped, but mild wound sepsis occurred after the second operation that improved with repeated dressings and broad-spectrum antibiotics. The third patient with acute large bowel obstruction underwent an emergency surgical exploration and primary left colectomy with intraoperative colonic lavage, as the tumor was resectable with no signs of intraabdominal metastasis according to abdominal CT and operative findings. The patient passed the first 5 days uneventfully, then developed persistent fever. The drain was dry until that time, and the wound was clean. Abdominal US revealed a large intraabdominal collection. Re-exploration showed severe leakage at the anastomotic line. Defunctioning ileostomy was performed to control severe sepsis, unfortunately the patient died 28 days postoperatively due to septic shock and multiorgan failure.

Discussion. Adult intussusception is a rare entity encountered by surgeons. It differs from intussusception in children in various aspects, regarding etiology, clinical presentation, diagnostic approach, and management.[2,7,8] Intussusception has been classified into 4 categories according to the site of origin, such as enteric, ileocolic, ileocecal, and colocolic.[9] Yakan et al[10] who evaluated 20 cases of AI found that most cases were in the small intestine (85%). The most common site of AI occurs in the small and large bowels with a reported incidence of 90%, and the remaining 10% involves the stomach, or a surgically-created stoma.[11] Colo-anal intussusceptions are rare, and 50% are attributable to a malignant lesion.[12] The overall incidence of malignancy is 43-56%.[1,2] Colocolic intussusceptions secondary to carcinoma occurs in up to 43-80% of the cases.[2,13] The mean age for AI in our study is 46.9 years, while in another study it was reported as 54.4 years.[2] The mean age for benign cases as reported by some authors is 44 years as opposed to 60 years for malignancy-related cases.[13] This is comparable with our results (44.4 years as opposed to 61.5 years for cases attributed to malignant lesions).

The lead points for intussusceptions are attributable to benign, malignant, or idiopathic causes.[7,11] In a review by Felix et al[14] tumor-related intussusceptions were noted in 63% of cases. Primary or secondary malignant lesions may account for 6-30% of all cases. In our series, benign lesions were detected in 78.6%, while malignancy was found only in 14.2%. The clinical presentation of intussusception is often chronic and most patients present with non-specific symptoms. Abdominal pain is the most common symptom followed by vomiting and nausea.[2,15] The clinical findings are variable: acute

intestinal obstruction is not common, and most patients present with subacute, chronic, or intermittent symptoms in adults.[16] In our series, recurrent episodes of abdominal pain were the most common symptom associated sometimes with vomiting, rectal bleeding, and signs of bowel obstruction.

Chronic AI requires a high index of suspicion for diagnosis as it provides vague, and often transient findings. Although the correct diagnosis is often based on intraoperative findings, modern imaging techniques like CT scan can be very helpful in precisely identifying these lesions preoperatively.[2,7,9,15] Failure to diagnose a fixed intussusception can lead to subsequent bowel ischemia, sepsis, and ultimately death. Plain abdominal radiographs are usually the first investigation performed, and the findings are non-specific. The classic findings on plain radiography are the presence of a large soft-tissue density from the intussusceptions. However, secondary findings, such as air-fluid levels, proximal bowel dilation, and collapsed distal bowel are more likely. In our cases, plain x-ray did not help, except in patients with intestinal obstruction.

In recent years, CT has become an important method for evaluating patients with non-specific abdominal complaints. The characteristics of intussusception on CT are an early "target mass" with enveloped, eccentrically located areas of low density.[17] Abdominal CT has been reported to be the most useful tool in the diagnosis of intestinal intussusception, and is superior to other contrast studies, US, or colonoscopy.[3,17] In recent series,[6,18,19] the reported diagnostic accuracy of CT scans was 58-100%.

In our study, CT scan confirmed the diagnosis of intussusception in all patients. It, therefore, appeared most useful in reaching the diagnosis, as well as detecting an unsuspected diagnosis as in the case of portal, superior mesenteric, and splenic vein thrombosis. Although contrast enema is seldom successful for the reduction of intussusception in adults, one of our patients demonstrated spontaneous reduction of an ileocecal intussusception during follow up CT scan with rectal contrast administration. Colonoscopy is also a useful tool for evaluating intussusception, especially when the presenting symptoms indicate a large bowel obstruction.[20,21] In our series, colonoscopy was performed in only 3 patients. The correct diagnosis was picked in only one patient that had left-sided colon cancer. In the patient with severe rectal bleeding, colonoscopy detected a sigmoid polyp, which was not the cause of bleeding, and missed the jejuno-jejunal intussusception with multiple polyps that was diagnosed after abdominal CT.

The treatment of AI is mostly surgical, as many cases have a definable pathological cause whether benign, or malignant. In our study, 85.7% had a definite cause. Weilbaecher et al[16] established the principle of resection without reduction whenever possible. Eisen et al[13] has reported that colonic lesions should not be reduced before resection as they most likely represent a primary adenocarcinoma. In our series, resection anastomosis after reduction was carried out in cases of small bowel intussusception to limit the extent of resection as all cases appeared benign, while resection without reduction was performed in all cases of large bowel intussusception as most of them looked malignant.

Recently, minimally invasive techniques have been introduced for the treatment of small or large bowel obstructions, specifically to the diagnosis and treatment of AI. Several laparoscopic small bowel resections secondary to intussusceptions have been reported.[22,23] In our study, we did not perform laparoscopy for management of these cases, however, in the future, we plan to use it according to our facilities in selected cases.

There was unavoidable information bias in our study as related to retrospective study design. The cases were recruited from only 2 medical centers in a short time interval, thus the limited number of cases. Therefore, future research will be needed including meta-analysis and larger retrospective studies to determine a better work up strategy, and a standard treatment of AI.

In conclusion, a high index of suspicion is necessary for reaching the diagnosis of AI as the symptoms are non-specific and intermittent. The CT scan has a major role in the diagnosis of this problem, especially in patients with undiagnosed abdominal pain. Surgical treatment with resection is the optimal treatment option.

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