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## Original article

## Intestinal invagination in adults: Presentation of a case and a review of the Spanish literature

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## A B S T R A C T

**Introduction:** Intestinal invagination in the adult is an uncommon condition, often manifested by non-specific chronic or sub-acute symptoms. In the majority of occasions there is an organic lesion. There are currently no large patient series published in the literature to help define the management of these patients.

**Material and method:** A review of case series published in the Spanish literature. A data base of patients over 15 years old was designed. Data was extracted from national clinical cases using Internet resources. Our own recent clinical case is added.

**Results:** A series of 30 adults with intestinal invagination was obtained (29 cases from the review and 1 own). The median age was 45 years (19–84 years) and 17/30 (57%) were males. A total of 27/30 patients had abdominal pain and 8/30 (28%) cases had established intestinal obstruction. The preoperative diagnosis of invagination was made in 25/30 (83%) of patients. The invaginations were; enteroenteric, 61%; colocolic, 12%; enterocolic, 21%; and gastrojejunal, 6%. A total of 85% of the invaginations were associated with a proliferative lesion and 43% of the latter were malignant.

**Conclusions:** The diagnosis of invagination in the adult is usually made preoperatively. There are no data to support intestinal resection without performing a reduction. Resection must be the norm and the presence of lymph nodes is no argument to perform large resections.

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## Invaginación intestinal en el adulto: presentación de un caso y revisión de la literatura médica española

## R E S U M E N

**Introducción:** La invaginación intestinal en el adulto es una entidad infrecuente, a menudo manifestada con síntomas inespecíficos crónicos o subagudos. En la mayoría de las ocasiones asocia una lesión orgánica. Actualmente no hay publicaciones de series amplias de pacientes que ayuden a definir el tratamiento de estos enfermos.

## Palabras clave:

Invaginación en adulto

Oclusión intestinal

Ecografía

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**Material y método:** Revisión de series de casos publicados en la literatura médica española. Se diseñó una base de datos de pacientes mayores de 15 años. Los datos se extrajeron de casos clínicos nacionales y se utilizaron recursos de Internet. Se añadió un caso clínico propio reciente.

**Resultados:** Se obtuvo una serie de 30 adultos con invaginación intestinal (29 de la revisión de casos y uno propio). La mediana de edad fue de 45 años (de 19 a 84); 17/30 (57%) fueron hombres. Presentaron dolor abdominal 27 pacientes y obstrucción intestinal establecida 8 casos (28%). El diagnóstico preoperatorio de invaginación se hizo en 25/30 pacientes (83%). Las invaginaciones fueron enteroentéricas (61%); colónicas (12%); enterocólicas (21%), y yeyunogástricas (6%). El 85% de las invaginaciones asoció una lesión proliferativa y el 43% de estas últimas fueron malignas.

**Conclusiones:** Habitualmente el diagnóstico de invaginación en el adulto se obtiene preoperatoriamente. No hay datos para apoyar la resección intestinal sin realizar reducción. La resección debe ser la norma, y la presencia de adenopatías no es un argumento para realizar resecciones amplias.

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## Introduction

Contrary to occurrence rates in children, intestinal invagination has a low occurrence rate in adults. When it occurs in the paediatric age, it usually manifests as occlusive symptoms, and the most frequent cause for intestinal obstruction is between 3 months and 6 years of age.<sup>1</sup> In adults, clinical signs correspond to symptoms of abdominal pain and are often associated with inflammatory biochemical symptoms; however, complete obstruction is not constant. There are no up-to-date publications with a wide range of case series on this disease. In a 1971 study which included 160 cases,<sup>2</sup> sub-acute and chronic types of this disease were described as more frequent, while a malignant form of the disease was found in 24% of intestinal invaginations and in 54% of colonic invaginations. Intestinal resection was recommended for all types of invagination in adults, without previous disinvagination. In 1997, Azar et al<sup>3</sup> carried out a retrospective study at the Massachusetts General Hospital with 58 patients over the age of 18, corresponding to a period of approximately 30 years (from 1964 to 1993). The preoperative diagnosis was properly carried out in 32% of the cases, and it was improperly performed with a higher frequency in benign small intestine invaginations (22% with a preoperative diagnosis of invagination). This study is widely cited in current articles in order to indicate the high number of cases without preoperative diagnosis linked with this condition. However, this is inaccurate. Following a new case of intestinal invagination, the cases published in the Spanish medical literature were reviewed, as was a database with an updated state of intestinal invagination in adults in Spain.

## Material and method

A bibliographical research on the Spanish medical literature was carried out using Dialnet ([www.dialnet.unirioja.es](http://www.dialnet.unirioja.es)), a dissemination portal for the Spanish scientific production,

the Elsevier database ([www.elsevier.es](http://www.elsevier.es)) and the Arán Ediciones database ([www.grupoaran.com/WEBN/Buscador.asp](http://www.grupoaran.com/WEBN/Buscador.asp)). Publications of clinical cases of the Association of Surgeons of Spain were also reviewed. The keywords used were “intestinal intussusception” and “intestinal invagination.” Patients younger than 15 years of age were excluded. The following data were included in the database: gender, age, abdominal pain, clinical intestinal obstruction, recurrent symptoms, leukocytosis, diagnostic method, immediate surgery, site of the invagination, number of invaginations in the same episode, type of surgery, disinvagination, and anatomopathological result. Patients treated in our services were included.

## Results

Twenty-three articles from the Spanish medical literature were selected concerning invagination in adults,<sup>4–26</sup> all of which were published between 2001 and 2008. All of the articles described the experience of a clinical case and discussed the subject, except for 2: one article included 2 cases<sup>4</sup> while another one consisted of 7 retrospective cases.<sup>5</sup> The latter article included a male patient whose clinical case served as a basis for another publicación.<sup>6</sup>

A case series of 30 adults was obtained, all of whom had intestinal invagination, an average age of 45 years (range, 19–84) and a gender distribution of 17 males (57%) and 13 females (43%). Abdominal pain was a primary symptom in 27 of the 30 patients, located in the epigastric and mesogastric area in the majority of the cases. However, an established clinical symptom of intestinal obstruction was only mentioned in 8 of the 30 patients (28%). For 17 patients, specific data were obtained concerning the presence of a mass during the abdominal palpation, which came back positive in only 4 of the patients. In 13 of the 20 patients (65%) there was an appearance of recurrent abdominal pain. Six of the 11 patients (55%) showed signs of leukocytosis in the complete blood

count. A preoperative diagnosis of intestinal invagination was made in 25 of the 30 patients (83%). Ultrasonography between the first examinations was mentioned in only 18 of the 30 patients. The diagnostic tests used can be found in Table 1.

Three of the patients had more than one invagination at different intestinal sites in the same episode: jejunojejunal

and ileocaecal,<sup>20</sup> ileoileal and ileocaecal,<sup>23</sup> and gastrojejunal and jejunojejunal.<sup>25</sup>

Deferred surgery was carried out in 13 patients, while the remaining 17 were immediately operated on following diagnosis or due to the symptomatology. Laparoscopic approach was carried out in 3 cases. The anatomic sites of the invaginations can be found in Table 2. Invagination operation was performed on 7 of the 9 patients so as to reduce it surgically. In the rest of the patients it was not specified in the clinical case report whether an invagination operation was performed. A type of resection was performed in all cases, except for 2. In the case of the lipoma diagnosed in the preoperative examination using ultrasonography and CT scan, it was decided to carry out disinvagination, caecotomy and removal of the submucosal tumour (in a 30-year old female). The other case without resection concerned double gastrojejunal and jejunojejunal invagination. At the moment of intervention there was a spontaneous regression of the stomach, followed by intestinal disinvagination without witnessing the cause. The anatomopathological findings can be found in Table 3.

**Table 1 – Diagnostic tests for the preoperative diagnosis of invagination**

	Diagnosis	Without diagnosis
Abdominal ultrasonography	4	–
CT scan	8	1 <sup>17</sup>
Abdominal ultrasonography and CT scan	3	–
Endoscopy and CT scan	1	14
Opaque enema	2	–
Colonoscopy	1	–
Abdominal ultrasonography and intestinal barium x-ray	1	–
EGD	–	1 <sup>9</sup>
Abdominal ultrasonography, intestinal and intestinal barium x-ray	1 <sup>26</sup>	–
Combination of tests	4 <sup>a</sup>	11 <sup>1b</sup>
Laparotomy	–	1 <sup>5</sup>

CT scan indicates computed tomography.  
<sup>a</sup>Ultrasonography, opaque enema, CT scan, and colonoscopy.  
<sup>b</sup>Ultrasonography, CT scan, sigmoidoscopy, and intestinal barium x-ray.

**Table 2 – Distribution of intestinal invaginations**

Enteroenteric	20/33 (60.6%)
Jejunojejunal	9
Ileoileal	10
Jejunoileal	1
Colocolic	4/33 (12.1%)
Colocolic	4
Enterocolic	7/33 (21.2%)
Ileocolic	4
Ileocaecal	3
Gastrojejunal	2/33 (6%)

## Discussion

Intestinal invagination in adults appears with subtlety. Contrary to the medical literature, it does not only appear as the cause for intestinal obstruction in adults. This incongruous high obstruction rate comes from data taken from historical case series. One of the most cited research papers is the retrospective study by Begos et al,<sup>27</sup> which included 13 patients from 1981 to 1994, and where 75% of the patients had symptoms of intestinal occlusion. Following the analysis carried out in our study, we can state that the established clinical occlusion is not frequent, contrary to dilatation of the intestinal loops in x-rays.

The presence of recurrent abdominal pain in the anamnesis is frequent. The presence of leukocytosis and an increase in C-reactive protein is also common, thus establishing a differential diagnosis of appendicitis with, above all, pain in the right iliac fossa or palpation of a painful mass. Ultrasonography is crucial in these cases.

**Table 3 – Anatomopathological findings**

Benign (n=16)		Malign (n=12)			
		Primary		Secondary	
Lipoma	5	Colon ADC	2	Metastasis of choriocarcinoma	1
Myofibroblastic tumour	2	Ileal ADC	1	Metastasis of melanoma	1
Inflammatory fibroid polyp	3	GIST	2	Metastasis of lung carcinoma	1
Peutz-Jeghers polyp	1	Low-grade mesenchymal tumour	1	Metastasis of sarcomatoid carcinoma	1
Hamartoma	1	Lymphoma	1		
Endometriosis	1	Multiple myeloma	1		
Lymphoid hyperplasia	2				
Enteritis caused by Anisakis	1				
ADC indicates adenocarcinoma; GIST, gastrointestinal stromal tumour.					

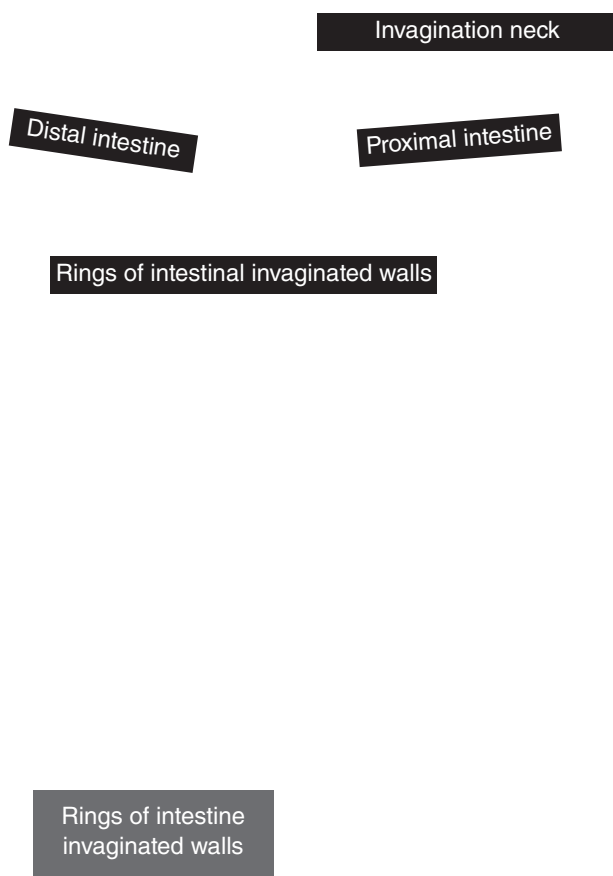
The palpable mass corresponds to invagination and does not make clear the size of the possible tumoral lesion causing the episode. Although not explicitly evident in the database, we were able to confirm weight loss and digestive blood loss as other signs related to invagination, without necessarily clarifying the presence of malignancy. Contrary to the published literature,<sup>3,4,7,27</sup> currently diagnosis is often carried out preoperatively. In this study, diagnosis was carried out prior to surgery in 25 of the 30 patients (83%). Preoperative diagnosis of the lesion causing invagination was carried out in 4 of the remaining 5 patients. Operative diagnosis was carried out in only one case of the 30 patients due to a lack of preoperative examinations of the acute abdomen. Concerning preoperative diagnostic tests, currently ultrasonography is frequently used because it is accessible in the majority of cases, including the consultation

of patients in ER. We do not believe that a preoperative CT scan is needed if ultrasonography reveals the right image (Figure). Other authors<sup>13</sup> consider a CT scan necessary to obtain comprehensive results given the high malignancy percentage associated with invagination. Nevertheless, in our opinion, this examination can be carried out postoperatively if the diagnosis is that of malignancy, which is normally associated with additional necessary examinations.

Regarding the site, invaginations are classified in enteroenteric (jejunojejunal, jejunoileal, and ileoileal), colocolic (actual or sigmoid-rectal), and enterocolic (ileocolic, when the end of the invagination is located in the ileum, and ileocaecal, when it is located in the caecum or in Bauhin's valve).<sup>28</sup> The site distribution is well established in the historical case series. In a 1954 study that reviewed 745 invagination cases diagnosed operatively, 39% were enteroenteric, 13% ileocolic, 21% ileocaecal, 17% colocolic, and 10% involved the stomach, the duodenum, or the stomata.<sup>29</sup> Distribution is similar in the Spanish case series analysed in our study, considering that the number of invaginations is lower (33 invaginations in 30 patients).

Surgery was different in 13 patients. The need for immediate surgery is infrequent. According to Stewart et al<sup>30</sup> and Spanish authors,<sup>4</sup> laparoscopy can be useful for this disease. The absence of palpation does not hinder laparoscopy, since resection is normal in adults. Refraining from carrying out disinvagination for oncological reasons is based on authors' opinions.<sup>3,13,17,27</sup> In our analysis this manoeuvre is explicitly mentioned in only 9 cases. In 7 of them there was a voluntary attempt for disinvagination. In our opinion, carrying out this manoeuvre depends on its influence on surgical exposure and resection, without significance in the spread of the tumour. If the presence of adenocarcinoma is suspected, the inclusion of lymphadenopathies will determine the extension of the resection. However, lymphadenopathies are usually inflammatory,<sup>6</sup> even in the presence of malignancy,<sup>13</sup> and not all malignant lesions require lymphadenectomy, for example, gastrointestinal stromal tumours or metastasis. If the small intestine is the affected area, the dilemma is not important, contrary to enterocolic or colocolic invaginations, where the vascular pedicles for the purposes of lymphadenectomy take over a larger area. Resection should be carried out sensibly in the context of the individual patient. The presence of lymphadenopathies should not be considered a case for adenocarcinoma.

In summary, contrary to historical case series, the diagnosis of intestinal invagination is only carried out prior to surgery. Frequently, surgery can be deferred or scheduled, which would facilitate the use of laparoscopy in this disease. There is no evidence to suggest refraining from disinvagination. The latter should be carried out if it facilitates the exposure conditions to perform intestinal resection. Concerning resection, weight loss, anorexia, and the presence of lymphadenopathies at the operative moment should not constitute reasons for widening it. Resection should be kept to a minimum while waiting for the anatomopathological result. Although associated with a high malignancy percentage, not all malignant neoplasms require lymphadenectomy, while the ganglia can only be reactive.



**Figure - A) Longitudinal ultrasonographic image of ileoileal invagination. The telescoping of a short segment of the ileum inside a more distal ileal segment can be seen in the longitudinal section. B) Transverse ultrasonographic image of ileoileal invagination. Transversal image where multiple concentric rings can be seen, representing the different layers of the intussusceptum wall, which invaginates into the intussusciens.**

## Conflicts of interest

The authors affirm that they have no conflicts of interest.

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