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CLINICAL CASE

# Intestinal intussusception secondary to myofibroblastic tumour in an elderly patient. Case report☆

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**KEYWORDS**
Tumour;
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**Abstract**
*Background:* Intussusception is defined as a segment of the gastrointestinal tract and mesentery within the lumen of an adjacent segment. It is a rare condition in adults that can occur anywhere in the gastrointestinal tract from the stomach to the rectum. Only 5% of all intussusceptions are presented in adults, and in 1–5% of all cases of intestinal obstruction. Inflammatory myofibroblastic tumour is rare, and is usually found in the lung, and rarely detected in some intestinal portions. It causes a variety of non-specific symptoms, with those that present as an intussusception being uncommon.
*Clinical case:* A female of 69 years with partial bowel obstruction secondary to intestinal intussusception due to an inflammatory myofibroblastic tumour, a rarely diagnosed condition and never published before.
*Discussion:* Inflammatory myofibroblastic tumours are rare, and in this case with an atypical presentation that was surgically resolved satisfactorily. These entities are difficult to diagnose, with histopathology giving the definitive diagnosis. A literature review was performed to gather recent information about their diagnosis and treatment.
*Conclusions:* Inflammatory myofibroblastic tumours require a high level of suspicion, as diagnosis prior to surgery is difficult. Surgery is considered the treatment of choice, requiring leaving free surgical edges to prevent recurrences.


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**PALABRAS CLAVE**
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Obstrucción intestinal

**Intususcepción intestinal secundaria a tumor miofibroblástico en un adulto mayor. Reporte de caso**

**Resumen**
*Antecedentes:* La intususcepción se define como la invaginación de un segmento del tracto gastrointestinal y su mesenterio, dentro de la luz de un segmento distal adyacente, puede ocurrir en cualquier lugar del tracto gastrointestinal, desde el estómago, hasta el recto. En adultos es rara, representa el 5% de todas las intususcepciones, y en el 1-5% de los casos de obstrucción intestinal. El tumor miofibroblástico inflamatorio es una entidad patológica rara, generalmente benigna, con predominio en el pulmón, y rara vez se detecta a nivel intestinal, lo cual ocasiona una variedad de síntomas inespecíficos, por lo que es poco frecuente que se presente como una intususcepción.
*Caso clínico:* Mujer de 69 años, con cuadro clínico de suboclusión intestinal secundaria a intususcepción, por un tumor miofibroblástico inflamatorio, rara vez diagnosticado y rara vez publicado.
*Discusión:* Los tumores miofibroblásticos inflamatorios son raros, en este caso con una presentación atípica, resuelta quirúrgicamente de forma satisfactoria. Es una entidad de difícil diagnóstico, el cual es definitivo con el estudio histopatológico. Con la finalidad de recopilar la información actual sobre su diagnóstico y tratamiento, se realizó una revisión bibliográfica.
*Conclusión:* Los tumores miofibroblásticos inflamatorios requieren un alto nivel de sospecha para realizar un diagnóstico previo a la cirugía. El tratamiento de elección ante este tipo de tumores es la cirugía, en la que se deben dejar bordes quirúrgicos libres para evitar recidivas.


## Background

Intussusception is defined as a segment of the gastrointestinal tract and mesentery within the lumen of an adjacent segment. The folded intestine slips into the adjacent intestine segment, propelled by peristalsis, with consequent intestinal obstruction and ischaemia.[1] Intussusception is a rare condition in an adult and can occur anywhere in the gastrointestinal tract from the stomach to the rectum. It represents approximately 1%–5% of all cases of intestinal obstruction. The mean age of presentation of intestinal intussusceptions in adults is 50, with a male/female ration of 1:1.3.[2] It has been established that this condition is found in less than one out of every 1300 abdominal operations and one out of every 100 patients operated on for intestinal occlusion. Mortality by intussusceptions in adults is 8.7% for benign lesions and 52.4% for the malignant variety.[2] Unlike children, in whom the majority of cases are idiopathic, intussusception in adults has an identifiable aetiology in 80%–90% of cases. Out of these, 50%–75% are due to benign diseases. Among the most common are the adhesions and Meckel's diverticulum. Other tumours include: lymphoid hyperplasia, lipomas, leiomyomas, hemangiomas and a minor proportion is idiopathic.[3]

Increased flaccidity of the intestinal wall facilitates intussusception.

Inflammatory myofibroblastic tumour is rare, is usually found in the lung, and rarely detected in the bowel. Some cases have been reported in the head of the pancreas and the duodenum. Although the aetiology of myofibroblastic tumours is not entirely known, the World Health Organisation classifies them as tumours of intermediate biological potential, since both local recurrence and metastases are possible, as is progression with malignant potential.[3,4]

The definitive diagnosis of this type of tumour is histopathological. Over 71% of inflammatory myofibroblastic tumours test positive for ALK1, which is a gene that is involved in the phenomena of vascular remodelling and angiogenesis. Its determination is genetic, in approximately 50% of cases it has a clonal rearrangement, related to the ALK locus, in chromosome 2p23, which is related to the neoplasic nature of the inflammatory myofibroblastic tumour.[5] ALK1 negative has been linked to elderly patients, and it also shows many nuclear pleomorphisms and atypical mitoses.[6,7] Since ALK1 positive is related to young patients, with a high recurrence, it is important to carry out the determination of ALK1, to establish disease prognosis.[8,9]

With regard to immunohistochemistry, these tumours test positive to vimetin without expression of CD 117 and CD32. The cells test positive to smooth cell actin with or without expression of desmin and S100 positive. All the previous ones are proteins expressed in the tumour tissue. They equally serve as a prognostic disease factor.[10,11]

Surgical resection is the treatment of choice for inflammatory myofibroblastic tumours.[12] Total surgical resection is associated with a recurrence under 10%. Chemotherapy is reserved for patients where surgical resection is morbid, impossible or incomplete. There is no evidence to show that chemotherapy would be effective as monotherapy, but it may play a role after surgery.[12,13] Radiotherapy is solely reserved for palliative care, with the objective