# Advanced adenocarcinoma of terminal ileum: an unusual neoplasm revealed by an unusual diagnostic tool

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Abstract. Background and aim: Terminal ileum adenocarcinoma is a rare tumour. Its incidence or prevalence among the other sites of gastro-intestinal tract is unknown, since it has been only sporadically described. Since contrast enhanced ultrasonography has been recently used to study bowel alterations in the course of neoplastic or inflammatory disorders, we report here a case of a rare tumour (terminal ileum poorly differentiated adenocarcinoma) in which the investigation played a pivotal role to obtain a defined diagnosis. Materials and methods (case report): Here we report the case of a 62 year old male patient. Due to intestinal occlusive symptoms and body weight decrease of about 8 Kg, he performed an abdominal computed tomography, intestinal magnetic resonance with double contrast medium, colonoscopy and contrast enhanced ultrasonography using a second generation medium. Results: In our case the peculiar aspect is that no arterial enhancement was observed and the finding remained unchanged for about 2.48 minutes as well as after a further administration of 1.5 ml of contrast medium. This aspect was not suggestive of an active inflammation such as Crohn's disease, where a marked contrast medium enhancement should be expected. Conclusions: At present it is too speculative to emphasize contrast enhanced ultrasonography as usefulness tool in the diagnosis of terminal ileum tumors. Nevertheless, our preliminary experience strongly encourages the diffusion of the method. (www.actabiomedica.it)

Key words: terminal ileum adenocarcinoma, contrast enhanced ultrasonography, Crohn's disease

## Introduction (Background and aim)

Terminal ileum adenocarcinoma is a rare tumour, with a still not well studied tumorigenesis process, usually presenting in an advanced stage. The clinical diagnosis is often difficult; surgery is the treatment of choice when feasible, while the chemotherapic approach is still not well codified (1). Its incidence or prevalence among the other sites of gastro-intestinal tract is unknown, since it has been only sporadically described often as a complication of Crohn's disease (2) or associated to carcinoid (3). Technological improvement of ultrasonographic instruments allowed to detail small bowel aspects such as the wall components, lumen aspect and peristalsis which show peculiar alterations in the course of inflammatory and neoplastic disorders (4). In the last years, intravenous contrast medium of second generation allowed detecting alterations of intestinal microcirculation such as hyperaemia and neoangiogenesis as well as detailing abscesses, fistulas or stenosis (5). Despite these undutiful progresses, scanty data are available about the differential diagnosis of inflammatory and neoplastic disorders of the small bowel. Since contrast enhanced ultrasonography has been recently used to study bowel alterations in the course of neoplastic or inflammatory disorders, we report here a case of a rare tumour (terminal ileum poorly differentiated adenocarcinoma) in which the investigation played a pivotal role to obtain a defined diagnosis showing some aspects which allowed to distinguish the tumour from Crohn's disease of the terminal ileum.

#### Materials and methods (case report)

Our male patient (62 years old) presented a past clinical history of a metabolic syndrome with obesity complicated by H. pylori related duodenal ulcer, reflux oesophagitis, recurrent thoracic pain, extrasystolic arrhythmias, chronic respiratory failure and prostatic hyperplasia. During a hospital admission in 2009 into an Emergency Surgery Unit for intestinal occlusive symptoms and body weight decrease of about 8 Kg, he performed an abdominal computed tomography that showed mild ascites, dilatation of jejunal and ileal loops, terminal ileum stenosis thus suggesting a clinical suspicion of Crohn's disease. After occlusion resolution, an intestinal magnetic resonance with double contrast medium (oral and intravenous) was performed and demonstrated and increase of the terminal ileum wall thickness and mild lymphoadenomegaly at the inferior mesenteric artery partition. The picture was again suggestive of an ileal inflammatory disease and the patient was admitted to our Unit of Gastroenterology. Laboratory investigations showed some neoplastic markers such as ferritin, Ca 19-9 and TPA increased value with a marked reduction of total proteins (5 g/dl) and the transabdominal ultrasonography showed a framework of liver steatosis, biliary sludge and enlarged intestinal loops surrounded by ascitic fluid with a marked reduction of peristaltic movements in the right inferior abdomen. Therefore, we performed a contrast enhanced ultrasonography using a second generation medium (Sonovue, Bracco - S. Donato - MI - Italy). The investigation confirmed the increase of the terminal ileum wall tickness (10 mm for a length of 20 cm), but after Sonovue administration (3.5 ml) no arterial enhancement was observed and the finding remained unchanged for about 2.48

minutes as well as after a further administration of 1.5 ml of contrast medium. This aspect was not suggestive of an active inflammation where a marked contrast medium enhancement should be expected (figure 1). A colonoscopy was finally performed and the instrument could not be introduced through the ileo-cecal valve. At this time, a laparatomy was performed showing omental diffusion of a tumour arising from terminal ileum. It histologically appeared to be a poorly differentiated adenocarcinoma (immunohistochemistry: strongly positive CDx2 diffusion marker, strongly positive cytokeratin 20, negative cytokeratin 7, negative antibodies to mesotelium). Further surgical procedures other than palliation to avoid occlusion and district exploration were not feasible.

## Results

Our case report shows some interesting aspects which could alert the reader:

- a. adenocarcinoma of the terminal ileum, even if very rare, needs to be considered when a picture suggesting a Crohn's disease of this area is observed at common invasive and non-invasive investigations such as colonoscopy and intestinal magnetic resonance;
- b.contrast enhanced ultrasonography appears as a promising approach in this case for the diagnosis achievement although the method has been pioneered only in this last period. In fact, in our case the peculiar aspect is that no arterial enhancement

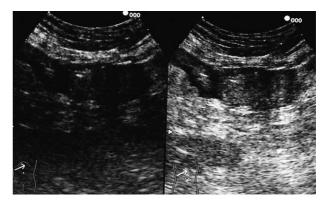


Figure 1. Left side: absence of contrast enhancement after 2'48" of Sonovue administration (late phase); right side: base-line ultrasonography

was seen and the finding remained unchanged for about 2.48 minutes as well as after a further administration of 1.5 ml of contrast medium. This aspect was not suggestive of an active inflammation such as Crohn's disease, where a marked contrast medium enhancement should be expected.

#### Conclusions

At present it is too speculative to emphasize contrast enhanced ultrasonography usefulness in the diagnosis of terminal ileum tumors; controlled multicentre studies are needed to well establish the possibilities and limits as well as to standardize the ultrasonographic parameters. Nevertheless, our preliminary experience strongly encourages the diffusion of the method.

#### References

 Manfredi S, Thiebot T, Henno S, Falize L, Bretagne JF, Meunier B. Complete response of an initially non-surgical adenocarcinoma of the duodenum to chemotherapy with the Folfox 4 regimen. J Gastrointest Surg 2009; 13 (12): 2309-13.

- Dossett LA, White LM, Welch DC, et al. Small bowel adenocarcinoma complicating Crohn's disease: case series and review of the literature. *Am Surg* 2007; 73 (11): 1181-7.
- Cioffi U, De Simone M, Ferrero S, Ciulla MM, Lemos A, Avesani EC. Synchronous adenocarcinoma and carcinoid tumor of the terminal ileum in a Crohn's disease patient. *BMC Cancer* 2005; 5: 157.
- Cammarota T, Sarno A, Robotti D, et al. US evaluation of patients affected by IBD: how to do it, methods and findings. *Eur J Radiol* 2009; 69 (3): 429-37.
- 5. Serra C, Menozzi G, Labate AM, et al. Ultrasound assessment of vascularization of the thickened terminal ileum wall in Crohn's disease patients using a low-mechanical index real-time scanning technique with a second generation ultrasound contrast agent. *Eur J Radiol* 2007; 62 (1): 114-21.

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Accepted: December 28th 2010

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