

Case Report

## Severe rectal prolapse and solitary rectal ulcer in a young anorectic woman with myxoid degeneration of visceral adipose tissue

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### Summary

Rectal prolapse is a disorder in which the rectum protrudes from the anal canal. Solitary rectal ulcer may coexist. Both conditions have been associated with chronic constipation and excessive straining during defecation. Rectal prolapse has been rarely reported in women suffering from anorexia nervosa. Lack of rectal support because of loss of ischioanal fat has been proposed as one of the possible mechanisms in this condition, together with chronic constipation and abuse of laxative. We report the case of an anorectic woman with a severe rectal prolapse and bleeding requiring urgent Altmeier's procedure. Surgery was complicated by dehiscence of the anastomosis and volvulus, requiring ileostomy and laciniae debridement. Pathological analysis of all the surgical samples taken from different abdominal sites highlighted changes in the visceral adipose tissue consisting in nodular aggregates of small adipocytes dispersed in a myxoid matrix surrounding blood vessels within abundant fibrosis. The morphologic features resemble those observed in primordial fetal fat and are comparable to those observed in cancer associated cachexia. The diffuse myxoid degeneration of visceral adipose tissue may play a role in the pathogenesis of rectal prolapse in patients with anorexia nervosa. Besides starvation, the mechanism sustaining myxoid degeneration of the adipose tissue is not entirely clear. Whenever possible improving nutritional and clinical conditions should be ideal before any surgical approach.

**Key words:** anorexia, myxoid degeneration, adipose tissue, rectal prolapse, fetal fat

### Introduction

Rectal prolapse is a disorder in which the rectum protrudes from the anal canal. It may be partial, involving the rectal mucosal only, or complete. It is more common in extreme ages, with a prevalence of 1% in adults over 65 years old. It is more frequent in women. The complete or partial protrusion of the rectum is accompanied by local signs and symptoms such as pain and bleeding during defecation, and gastrointestinal dysfunction<sup>1</sup>. Solitary rectal ulcer is often, but not always, documented in rectal prolapse. Both conditions have been associated with chronic constipation and excessive straining during defecation. In this setting, abnormal function of anal and pelvic floor musculature, associated with increased mobility of the rectum, may cause mucosal rectal prolapse that in turn

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### Conflict of interest

The Authors declare no conflict of interest.

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leads to stretching of the submucosal blood vessels with consequent localized ischemia and mucosal ulceration<sup>2</sup>.

Rectal prolapse has been rarely reported in women suffering from anorexia nervosa<sup>3</sup>. Lack of rectal support because of loss of ischiorectal fat has been proposed as one of the possible mechanisms in this condition, together with chronic constipation and abuse of laxatives. The causative role of chronic starvation in rectal prolapse is also underlined by its frequency in pediatric patients with acute diarrheal illness superimposed on malnutrition<sup>4</sup>.

Here we report the case of a 29-years-old woman with anorexia nervosa presenting with a severe rectal bleeding prolapse, requiring a surgical approach, which was followed by life threatening complications. Pathological analysis revealed a diffuse visceral adipose tissue degeneration in the setting of severe starvation.

## Case presentation

A 29-years-old woman was admitted to the emergency department because of abdominal pain accompanied by bleeding during defecation.

The woman had been suffering from anorexia nervosa restricting subtype and depression from the age of 16. The patient reported no episodes of binge eating or purging behaviors. She was cachectic, with a body mass index (BMI) of 14 kg/m<sup>2</sup>. Laboratory investigations showed severe anemia (HB 7.0 g/dl) associated with hypokalemia (2.1 mEq/L). Physical examination revealed a complete irreducible rectal prolapse (8 cm). A transanal echography excluded the presence of rectal lesions while an anorectal manometry showed an internal anal sphincter hypotrophy (not shown). A colonoscopy could not be performed because of electrolyte imbalance and bradycardia that surged after assumption of the laxative preparation. Due to the severity of the rectal prolapse and anemia, an Altmeier's surgical procedure was performed, with the packaging of a colon-anal anastomosis.

The proctocolectomy's gross examination showed a circumferential thickening of the rectal wall and mesorectum with massive dilatation of the lumen (8 cm diameter) (Fig. 1 A-C). At the level of anterior rectal wall, 5 cm from the anal verge, a well demarcated area of mucosal erosion was evident (Fig. 1B). Histologically it was characterized by ischemic-type necrosis overlying a markedly hypertrophic muscularis mucosae. Glandular crypts surrounding the erosion appear hyperplastic and vascular congestion was evident (Fig. 1D-E). Tunica muscularis was hypertroph-

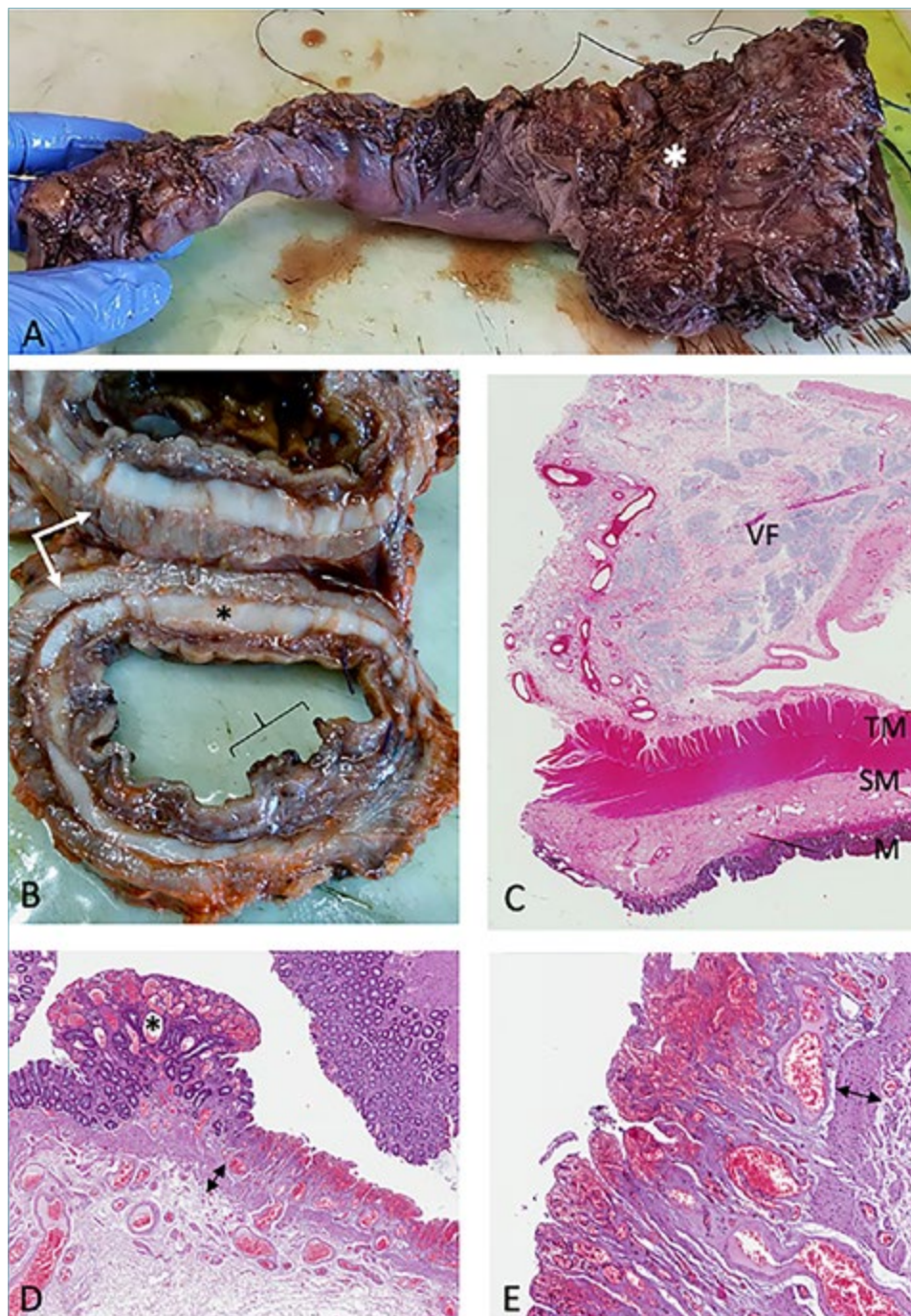
ic (Fig. 1C). According to the described morphologic features a diagnosis of solitary rectal ulcer associated with rectal prolapse was performed.

Besides rectal ulcer, a most striking morphologic feature was detected at the level of perivisceral fat, which was markedly thickened and presented a jelly consistency with a lobulated architecture consisting of nodules ranging in size from a few mm up to 10 mm (Fig. 1B-C). At histology nodules were characterized by a myxoid bluish matrix, surrounding vascular and nervous structures, with dispersed adipocytes showing multivacuolated cytoplasm and indented nuclei, reminiscent lipoblasts (Fig. 2A-B). The adipocyte nature of these cells was confirmed by immunohistochemical analysis for S100 (Fig. 2C). Nodules were surrounded by a loose fibrous tissue, as highlighted by Picro Sirius Red stain (Fig. 2D). Interestingly, the features resembled those of primordial fetal fat lobules (Fig. 2E)<sup>5</sup> and were reminiscent of a myxoid liposarcoma<sup>6</sup>. A tiny plexiform capillary vascular network was evident within the nodules in absence of the so called "chicken-wire pattern", which is usually evident in lipomatous malignant tumors (Fig. 2F). To completely rule out the hypothesis of a neoplasia, NMR was performed which showed thickening of the sigma wall and its meso in the absence of retroperitoneal masses suggestive of neoplasia. On the other hand, it revealed the presence of free abdominal air and ascites, pointing to a dehiscence of the colon-rectal anastomosis. Accordingly, an urgent ileostomy was performed, with a prophylactic appendicectomy. Cytologic analysis of peritoneal lavage confirmed the absence of neoplastic cells. Gross and microscopic analysis of the appendix showed similar features to that described for the rectum with myxoid appearance of visceral adipose tissue.

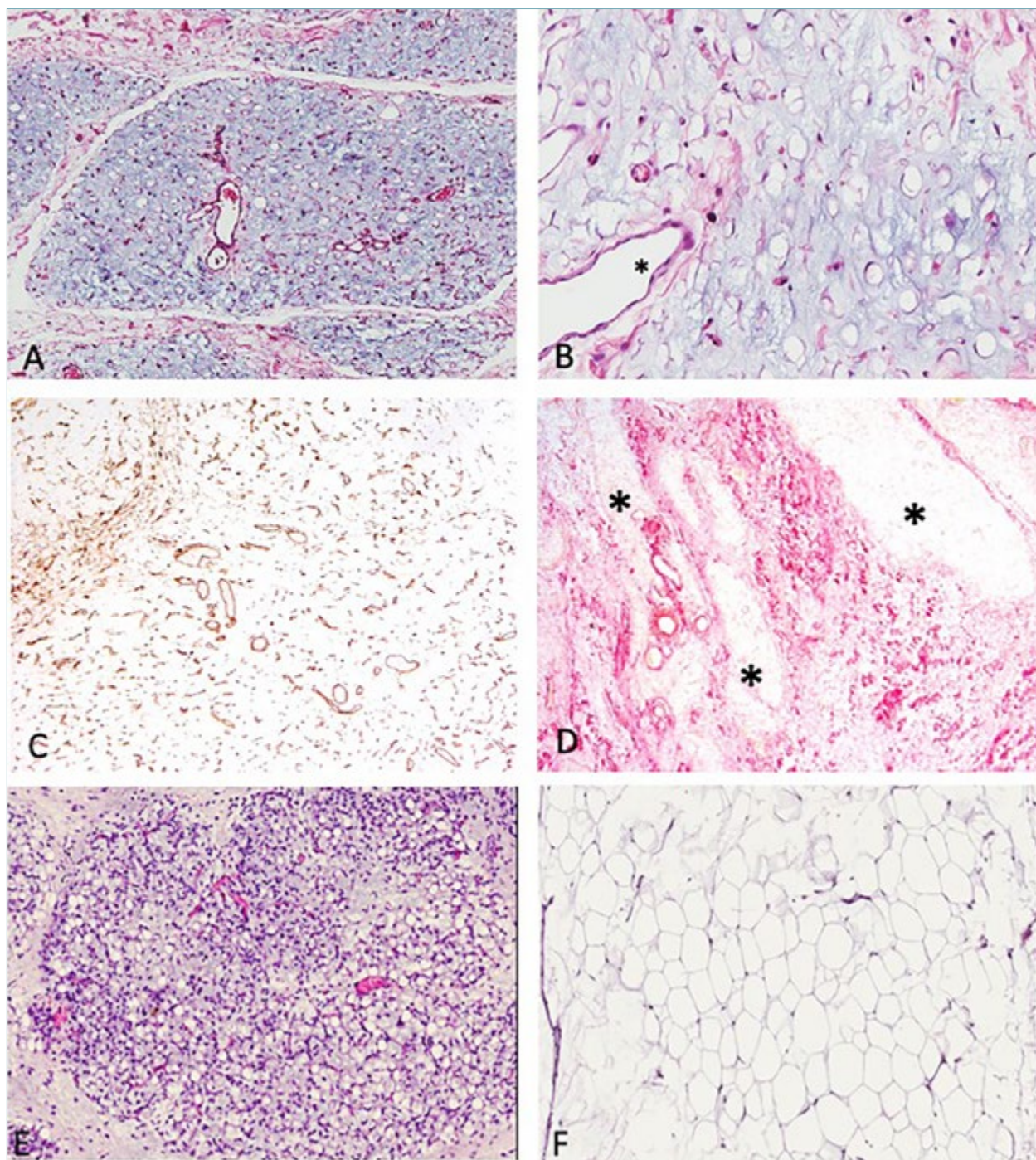
The post-operative period was characterized by a worsening of woman psychiatric symptoms. She presented profound depression, loss of appetite and dysphagia (BMI 12 kg/m<sup>2</sup>). A central catheter for nutrition was inserted and psychiatric therapy was optimized. After 10 days she developed an intestinal occlusion caused by adhesion bands with a volvulus requiring a third operation. Again, gross and histologic analysis of surgical samples revealed lobulated, myxoid-like visceral adipose tissue. Post-surgical recovery was characterized by improvement of psychiatric status and of her body weight (BMI 13 kg/m<sup>2</sup>).

## Discussion

Anorexia nervosa is a psychiatric disorder consisting in voluntary restriction of food, which leads to an ex-



**Figure 1.** Morphologic features of the gross intestine. (A) Proctocolectomy specimen showing bowel dilation, more evident in the distal portion of the rectum. Asterisk indicate the mesorectum. (B) Cross section of the rectum showing a markedly dilated lumen. Mucosa presents an area of erosion (bar graph), submucosa is unremarkable. Muscularis propria is thickened (asterisk). The perivisceral fat shows a nodular, gelatinous appearance (arrows). (C) Macrosection of the posterior rectal wall. Mucosa (M) and submucosa (SM) are unremarkable. Tunica muscularis (TM) is thickened. Visceral fat (VF) presents a lobular architecture with myxoid (bluish) nodules (Hematoxylin and eosin, original magnification 2X). (D) Mucosa from the anterior rectal wall showing superficial erosion and ischemic-like necrosis. Muscularis mucosae appears hypertrophic (arrow). Glandular crypts are hyperplastic (asterisk) (Hematoxylin and eosin, original magnification 10X). (E) Higher magnification showing mucosa ischemic-like necrosis with vascular congestion. Muscularis mucosae appears hypertrophic (arrow) (hematoxylin and eosin, original magnification 20X).



**Figure 2.** Microscopic features of visceral fat. (A) Myxoid nodule composed of adipocytes dispersed in a myxoid matrix. The nodule is organized around vascular structures (asterisk). (Hematoxylin and eosin, original magnification 10X). (B) At higher magnification adipocytes appear shrunken. They present a large cytoplasmic vacuole that displace the nucleus marginally. Vessel is highlighted by asterisk (Hematoxylin and eosin, original magnification 40X). (C) Immunohistochemical stain for S100 highlights adipocytes within a nodule (original magnification (Original magnification, 20X). (D) Picro Sirius Red stain shows abundant fibrous tissue surrounding myxoid fat nodules (highlighted by asterisks) (original magnification, 5X). (E) Fetal fetal lobule at 21 weeks' gestation. Adipose tissue sample has been obtained at autopsy in a case of therapeutic abortion (Hematoxylin and eosin, original magnification, 10X). Note the smaller size of adipocytes as compared to mature visceral adipose tissue (insert). (F) Immunohistochemical stain for CD34 shows a tiny plexiform capillary vascular network within a myxoid visceral fat nodule (original magnification, 20X).

treme loss of body weight<sup>7</sup>. It is a widespread pathology, most frequent in women. Its prevalence is reported to be between 1 and 4% and represents the psychiatric illness with the highest mortality rate, related both to the cachectic condition and to systemic complications involving the endocrine, cardiovascular, musculoskeletal, hematopoietic, and reproductive the system. Particularly frequent among woman with anorexia is gastrointestinal (GI) dysfunction, consisting of reduced GI tract motility, gastric dilation, constipation and, rarely, rectal prolapse<sup>8</sup>. The latter may result from several different factors, including chronic constipation and pelvic floor muscle weakness caused by malnutrition.

Here, we report a case of a young anorexic woman with a rare presentation of complete rectal bleeding prolapse associated with solitary rectal ulcer. According with the severity of rectal prolapse and anemia, a surgical approach was pursued, which was followed by a sequela of post-operative complications. Pathological analysis of all the surgical samples taken from different abdominal sites highlighted changes in the adipose tissue consisting in nodular aggregates of small adipocytes dispersed in a myxoid matrix surrounding blood vessels. Similar morphologic changes have been described in patients with cancer-associated cachexia, in which decrease in adipocytes size is paralleled by increase of collagen fibers and inflammatory cell infiltration in the extracellular matrix<sup>9</sup>. Experimental models attributed cachexia-related adipose tissue changes to increased lipolysis, reduction of lipogenesis and impairment of adipose tissue development and turnover<sup>10</sup>.

Interestingly, the morphological changes described in our case closely resemble those observed in primordial fetal fat. Fat lobules are, in fact, the earliest structures identified before the appearance of typical adipocytes in the fetus and are composed of mesenchymal cells derived from progenitors residing in the wall compartment of primordial proliferating vessels<sup>11</sup>. Around 14-24 weeks' gestation the appearance of cytoplasmatic lipid droplets convert mesenchymal cells to multivacuolated lipoblasts<sup>12</sup>. These cells, in the presence of systemic stimuli, can proliferate, migrate, and terminally differentiate<sup>13</sup>. Altogether, these histologic features mimic lipomatous or even epithelial tumors, thus requiring a thorough clinical and anatomopathological differential diagnosis<sup>14</sup>. The evidence that myxoid degeneration of visceral adipose tissue diffusely involved different portion of the GI tract, including the appendix, helped to rule out this hypothesis in our case.

In bone marrow, myxoid degeneration of adipose tissue is a well-known phenomenon associated with

starvation and is considered an important cause of refractory peripheral pancytopenia in anorexic patients<sup>15</sup>. Although the exact pathogenesis is still not known, it is possible that fat mobilization followed by the deposition of hyaluronic acid interferes with the hematopoietic microenvironment leading to the suppression of hematopoiesis. In line with this hypothesis, nutritional support has been shown to restore bone marrow function only when associated with hematopoietic growth factors<sup>16</sup>.

We may speculate that the diffuse degeneration of visceral adipose tissue contributed not only to rectal prolapse, but also to the postsurgical complications our patient experienced. In fact, by reducing the ability of mesenteries to fix the gross intestine to the posterior abdominal wall, it might lead to great mobility of sigmoid colon and rectum, favoring both prolapse and volvulus. On the other hand, rectal wall thickening because of myxoid degeneration of visceral fat might have induced poor visceral motility, another possible risk factor for rectal prolapse. Similarly, the surgical dehiscence might be attributed to the weakness of the intestinal wall caused by the adipose tissue degeneration.

The pathophysiological mechanism sustaining adipose tissue degeneration in anorexia nervosa is not entirely clear. Beyond starvation and malnutrition, cytokine storm, that is known to occur in anorectic patients, might play a role in the described changes<sup>17</sup>.

## Conclusion

Perivisceral adipose tissue degeneration may represent a morphologic substrate predisposing to rectal prolapse in anorexic patients. A surgical approach in the most severe cases of rectal prolapse is sometimes necessary. Surgery in emergency carries the risk of severe complications due to the extreme weakness of tissues. Whenever possible improving nutritional and clinical conditions should be ideal before any surgical approach.

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