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ONLINE CASE REPORT

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Transmural peritoneal adenomatoid tumour in the ileocaecal region causing massive haemoperitoneum and low gastrointestinal bleeding: differential diagnosis with capillary haemangiomas

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ABSTRACT

Peritoneal adenomatoid tumours are rare benign neoplasms originating from mesothelial cells. We present a case of peritoneal adenomatoid tumour penetrating into the bowel wall and causing massive intra- and extraluminal bleeding.

KEYWORDS

Peritoneal angiomatoid tumour – Massive low gastrointestinal bleeding – Massive hemoperitoneum

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Peritoneal adenomatoid tumours are rare benign neoplasms originating from mesothelial cells.[1] They usually appear between 25 and 55 years of age.[2] Their aetiology has not been established. Few patients present symptomatology related to the tumour and they are incidentally discovered during radiological examinations, surgery or postmortem examinations.[3] We present a case of a peritoneal adenomatoid tumour penetrating into the bowel wall and causing massive intra- and extraluminal bleeding.

Case history

A 48-year-old woman, with personal history of enolic liver cirrhosis, came to the emergency department of our institution complaining of massive rectal bleeding during the previous 24 hours. Physical examination revealed mucocutaneous pallor, tachycardia and hypotension at 80/40mmHg. The abdomen was slightly distended without tenderness. Rectal examination revealed massive rectal bleeding without evidence of a possible cause. Laboratory data showed haemoglobin at 6g/dl and haemostatic disorder (Quick rate 36% and activated partial thromboplastin time of 52 seconds). The patient was transferred to the intensive care unit because of haemodynamic instability. Resuscitation was started with a crystalloid infusion and transfusion of 4 units of packed red blood cells. A colonoscopy observed the whole colon full of fresh red blood, without evidence of bleeding point. The patient remained haemodynamically unstable and an exploratory laparotomy was performed revealing a 6l haemoperitoneum and the whole colon full of blood. A tortuous angiomatoid transmural lesion was located in the ileocaecal region with active intra-and extraluminal bleeding. A right hemicolectomy was performed. Postoperatively, the haemodynamic instability deceased 6 hours after surgery.

The histopathological study revealed a lesion composed of both solid and tubular regions, the latter penetrating from the serosal to the mucosal layer through the muscularis propria. The tubules were lined with a single layer of flattened cells, suggestive of endothelial cells. The angiomatoid architecture led to the initial suspicion of hemangioma (Figs 1 and 2). To confirm the diagnosis, immunohistochemical staining was performed, observing CD31 (endothelial cell marker) negativity (Fig 3) and calretinin (mesothelial cell marker) positivity (Fig 4), achieving the correct diagnosis of a peritoneal adenomatoid tumour.

Discussion

Peritoneal adenomatoid tumours usually involve the genital tract of both sexes, appearing more frequently among males. Among women, adenomatoid tumours are more commonly located in the myometrium, fallopian tubes, paraovarian connective tissue and, rarely, in the ovaries. Among men, they usually appear in the inferior pole of the epididymis, ejaculatory duct, spermatic cord, tunica albuginea, tunica vaginalis, testicular parenchyma and prostate. Other reported locations of adenomatoid tumours are the omentum, mesentery, pancreas, liver, bladder, mediastinum, pleura, heart and adrenal glands.[1,3–5] Although these tumours are thought to be of mesothelial origin, rare extragenital adenomatoid tumors have been reported and there are fewer than

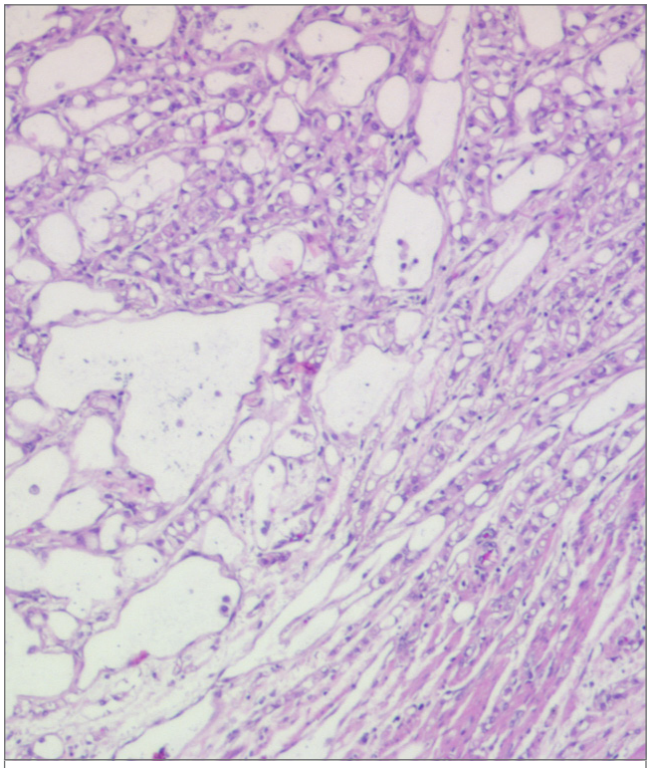

Figure 1 Stained with haematoxylin and eosin; 4x magnification: papillary formations penetrating into muscular layer

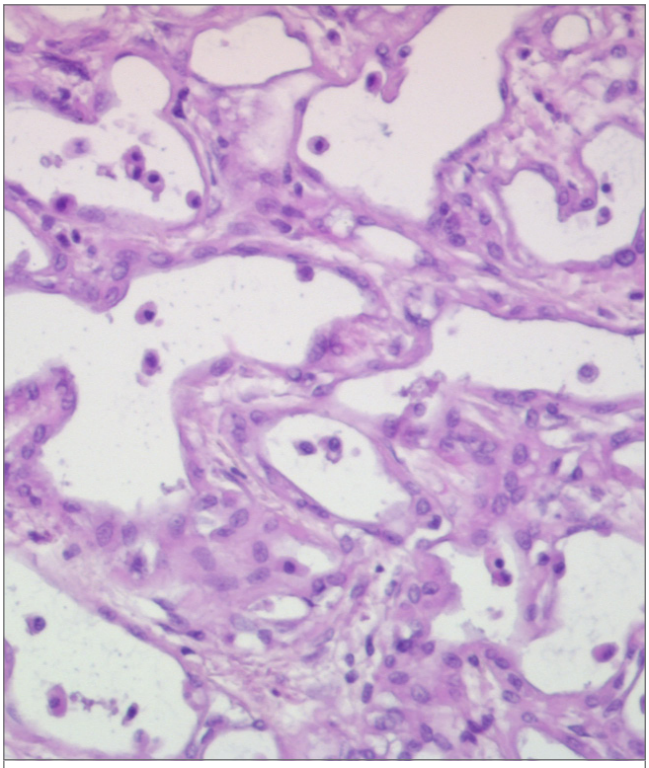

Figure 2 Stained with haematoxylin and eosin; 20x magnification: papillary formations lined by flattened cells, suggestive of capillary haemangioma

20 cases reported in the literature that have developed on the serosal surface. (In the case we present, the tumour is located on the serosal surface of the ileocaecal transition.) The mesothelial histogenesis still cannot explain the tendency for such tumours to occur much more frequently in the genital tract and more rarely on the serosal surface.[3,4,6,7]

Most peritoneal adenomatoid tumours are asymptomatic. When present, signs and symptoms include abdominal pain, weight loss, anorexia, nausea, ascites or a palpable pelvic mass.[1]

Usually, surgical resection is the treatment of choice for peritoneal adenomatoid tumours. Although benign, adenomatoid tumours are a source of great concern due to the differential diagnosis of malignant entities, with the obvious therapeutic implications.[8] Adenomatoid tumours may present local recurrence but they have no potential for malignant transformation.[1,5]

Macroscopically, peritoneal adenomatoid tumours are usually solitary, well circumscribed but non-encapsulated, yellowish-gray coloured polypoid or nodular small lesions (2cm or less), sometimes with small cysts.[2] In contrast, the gross aspect of the lesion in our patient resembled an angiomatoid lesion.

The 'adenomatoid' designation was introduced by Golden and Ash in 1945 because the cells are arranged in a cohesive manner, forming tubules and canaliculi.[1,2,4] Microscopically, adenomatoid tumours are typically composed of acini, tubules and anastomosing channels lined with a single layer of flattened or epithelioid cells, demonstrating little nuclear atypia or mitotic activity.[6] These tubules and channels may resemble vascular structures, especially if they contain red blood cells as in our case; the lesion was therefore initially considered to be angiomatoid, consistent when considering the clinical manifestations of the patient (rectal bleeding).

In the differential diagnosis of adenomatoid tumours, an immunohistochemical staining is essential. Tumoural cells are immunopositive for broad-spectrum cytokeratins, epithelial membrane antigen, cytokeratin 5/6, calretinin, HBME-1, WT1, D2-40 and vimentin, and immunonegative for CD31 and CD34.[3,4,6,7] CD31 allows the immunohistochemical staining of endothelial cells; in our patient the immunonegativity for CD31 discarded the diagnosis of haemangioma. Nevertheless, the positive staining for calretinin indicated the correct diagnosis of a peritoneal adenomatoid tumour.

It is especially remarkable that in our case the tubules of mesothelial origin penetrate through the muscular propria up to the mucosal layer. To our knowledge, this penetration of adenomatoid tumoural cells in the bowel wall has not been previously described and we believe it conditioned the intraperitoneal and intraluminal bleeding.

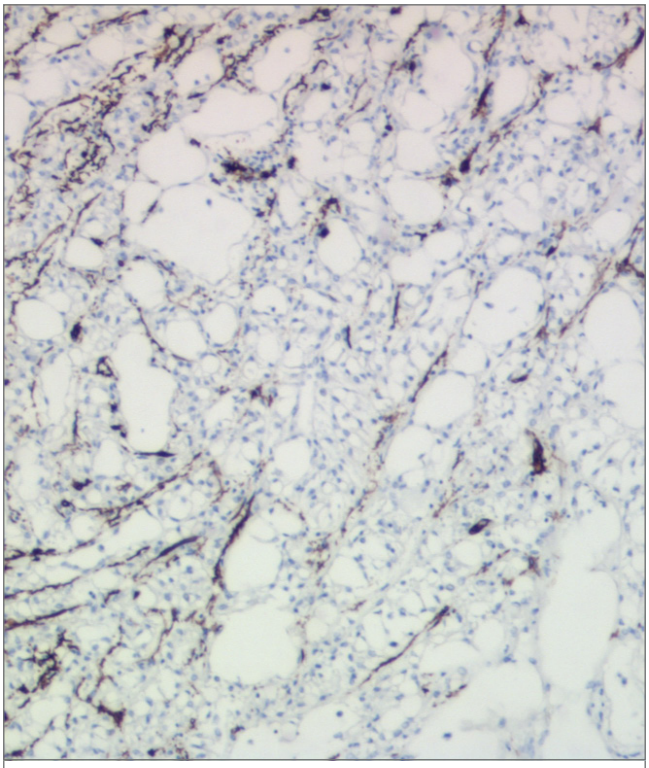

Figure 3 Immunohistochemical staining with CD31; 20x magnification: papillary formations are CD31 negative

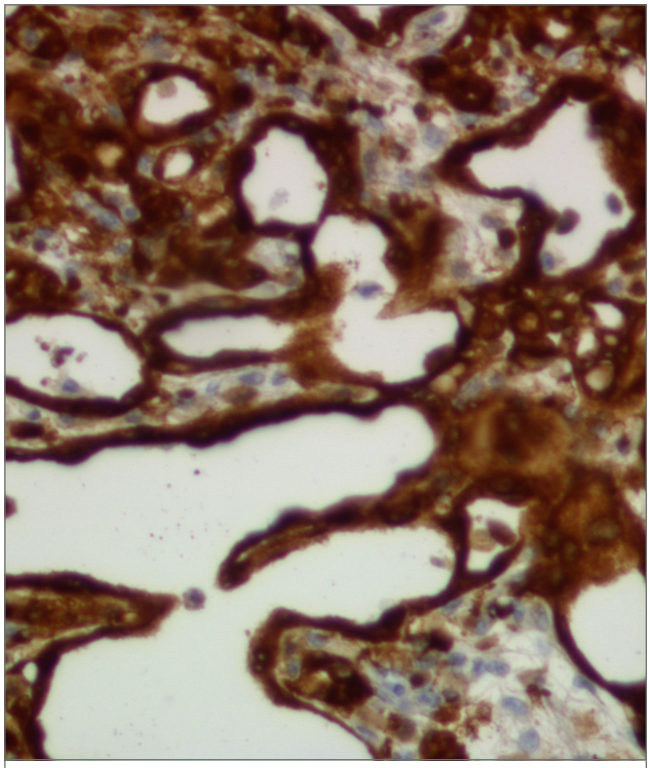

Figure 4 Immunohistochemical staining with calretinin; 20x magnification: papillary formations are calretinin positive

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