

CASE REPORT

Mesenteric panniculitis, a rare cause of acute surgical abdomen in children. Case report and literature review

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Abstract

Mesenteric panniculitis appears as a less known benign condition due to its infrequency and not fully elucidated etiology; currently, less than 20 cases in children have been reported. It is characterized by a fibrotic and inflammatory transformation of the mesentery fat of the small intestine and, less commonly, of the colon. The 8-year-old patient whose case we report presented in our Service complaining of acute digestive symptoms. A causality relation with the small yet frequent injuries the child suffered at the abdominal level during his sports practice may be hypothesized, although this correlation cannot be proven. Laboratory test values are usually within the normal range in such patients, yet the inflammatory values are sometimes high, just as in our case. The imagistic diagnostic workup detected a solid mass at the level of the hepatic colic flexure, with a non-homogenous structure, with fine contrast uptake in the walls; perilesional fatty infiltration with "misty mesentery" appearance; infracentimetric adenopathies located in the mesentery root. The tumor was surgically excised and intestinal anastomosis was performed. Achieving the final pathological diagnosis of mesenteric panniculitis was a difficult task, as it required several differential diagnoses, by ruling out a local vasculitis process and an idiopathic inflammatory Crohn's-like disease. The patient's post-operative evolution was positive. The follow-up examinations at one month, six months, one year and two years showed a good general condition, a good nutrition state and clinical-paraclinical test results within normal values. A significant association of mesenteric panniculitis to other malignancies, as well as a predisposition of these patients to the subsequent occurrence of neoplasms has been noted in adults. As these findings have not yet been proven in children, due to the small number of cases and the absence of prospective studies, long-term monitoring is an absolute must.

Keywords: mesenteric panniculitis, mesenteric lipodystrophy, abdominal pain, children.

Introduction

Mesenteric panniculitis is also known as mesenteric lipodystrophy. It is a benign condition characterized by the fibrotic and inflammatory transformation of mesentery fat [1]. Little is known about this condition, as only 17 of the 217 cases reported between 1965 and 2010 were children [2]. The first case was reported in 1924, although studies conducted on large groups of patients examined by computed tomography (CT), or on 700 necropsies, suggest the fact that the disease is underdiagnosed, as it really occurs in 0.6–2.5% of the population [3–7]. An interesting study carried out by Nicholson *et al.* revealed that only one out of 10 surgeons and one out of 12 radiologists in the institution where he worked were familiar with this condition [5]. Most of the cases are reported in the USA, France and Japan, in adults aged 50 and over, with a median age of 65 years and a male/female ratio of 1.8–3/1. The diagnosis of the condition is extremely rare in children, probably due to the small amount of fat in their mesentery [8–10]. The disease impairs the mesentery of the small intestine in over 90% of the cases, and, more rarely, the colonic, omental or pancreatic fat [4, 11].

Aim

We report hereunder the case of an 8-year-old child with a postoperative diagnosis of mesenteric panniculitis in the hepatic flexure in the colon. The patient had no history of disease or injuries and presented to our Hospital with symptoms of acute surgical abdomen.

Case presentation

The 8-year-old male patient, P.M.O., was brought to the Emergency Room of the "St. Mary" Emergency Children's Hospital, Iași, Romania, on November 19, 2014, complaining of a poor general condition, three-day-old colicky and increasingly severe abdominal pain accompanied by lack of appetite, food vomiting and fever. The child had judo practice twice a week and during the past two months suffered multiple minor abdominal traumas. According to the general practitioner's recommendations, he was given antibiotic and anti-inflammatory medication for two days, yet his general condition did not improve. The biological tests on hospitalization revealed slight anemia, namely hemoglobin (Hb) 11.3 g/dL [normal values (NVs): 11.5–14.5 g/dL], white blood cell count

(WBC) within normal limits, high inflammatory values: erythrocytes sedimentation rate (ESR) 61 mm/1 h (NVs: 0–12 mm/1 h), fibrinogen (Fb) 827.5 mg/dL (NVs: 180–400 mg/dL), C-reactive protein (CRP) 124.66 mg/L (NVs: 0–5 mg/L). The abdominal ultrasound scan revealed a concentric tumoral mass under the liver, with hypoechogenic layers alternating with hyperechogenic ones, with a 0.67 cm thick wall, which seemed to belong to the ascending colon; mesentery thickening around the detected lesion; infracentimetric mesenteric adenitis present; no other abdominal alterations. The thoracic-abdominal X-ray revealed the presence of a hydroaeric level in the right iliac fossa. The abdominal CT scan detected a cockade-like 4.3/6.3/7.3 cm mass under the liver, with a non-homogenous structure, on the topography of the hepatic flexure in the colon, with fine contrast uptake in the walls, which involved the second and third segments of the duodenum (Figure 1, a and b); perilesional fatty infiltration with “misty mesentery” appearance; infracentimetric adenopathies located in the mesentery root; no other pathological changes in the abdomen.

The patient was hospitalized in the Department of Pediatric Surgery, the File No. 60444, and after 10 hours, a surgical procedure was undertaken, namely supra-umbilical median laparotomy, in order to explore the peritoneal cavity. A hard 5/6/8 cm infiltrative tumoral mass with blurred borders and adhering omentum was detected in the hepatic flexure of the colon, together with several adenopathies at the foot of the mesocolon. The procedure consisted in the resection of the adhering omentum, of the tumoral mass with 5–6 cm of ascending and transverse colon (margins of safety) and of the related mesocolon. A few ganglia at the foot of the mesocolon were also sampled. Termino-terminal anastomosis and peritoneal drainage were then performed. The patient's post-operative evolution was positive and he was discharged five days after surgery. The follow-up examinations showed a good general condition, a good nutrition state and clinical-paraclinical test results within normal values. The inflammatory values were normal again on the next follow-up examinations (one month, six months and one year after the surgery), whereas the α -fetoprotein and beta-human chorionic gonadotropin (β -HCG) were normal both on hospitalization and on the subsequent performed checks (six months, one year and two years).

The final histopathological diagnosis of mesenteric panniculitis was set by corroborating the findings of three different specialized centers, and required a differential diagnosis with a local vasculitis process and a Crohn's-like idiopathic inflammatory disease.

Macroscopic exam revealed a 22 cm long colon fragment showing at 7.5 cm from the fragment end, at the level of the mesenteric adipose tissue, an imprecisely delimited area with increased consistency, gray and white coloring, with hemorrhagic zones of 5.5 cm. The subjacent mucosa was edematous, ulcerated, fixed on the tumor plan, and the serosa presented with hemorrhagic suffusions (Figures 2 and 3). The extemporaneous exam (frozen sections) and the smear showed an intestinal wall infiltrated with mature and immature lymphoid cells and granulocytes in different stages of maturation.

On paraffin sections, the sections of the resected colon ends showed a discrete inflammatory infiltrate in the lamina propria of the mucosa, congestion in submucosa and perivascular lymphocytes. The most intense changes were observed at serous level: abundant inflammatory cells with perivascular, diffuse and nodular disposition and fatty infiltration in the mesocolon (Figures 4–6). At this level, we also noticed images of obliterant endarteritis, perineuritis and focal vasculitis (Figures 7–9) associated with hemorrhagic areas (Figures 10 and 11) and foci of ischemic necrosis with dystrophic calcifications (Figure 12). In some histological sections, we observed foci of leukocyte exudate in the muscular wall affecting the myenteric plexuses and nerves, and in the peritoneal serosa together with granulation tissue rich in newly formed capillaries, fibroblasts, collagen and inflammatory cells (Figures 13 and 14). Eight fragments taken from the lesion revealed small ulceration of the colonic mucosa epithelium with increased polymorphic inflammatory infiltrate in the mucosal layer and also with lymphoid follicle hyperplasia. The lymph nodes showed hyperplasia of lymphoid follicles, increased stasis and histiocytosis in the lymphatic sinuses (Figures 15 and 16). The immunohistochemical (IHC) stainings allowed the differentiation of inflammatory cells. In the submucosal layer and peritoneal serosa, the polymorph inflammatory infiltrate showed CD3+, CD20+, myeloperoxidase (MPO)+, or CD68+ (Figures 17–20).

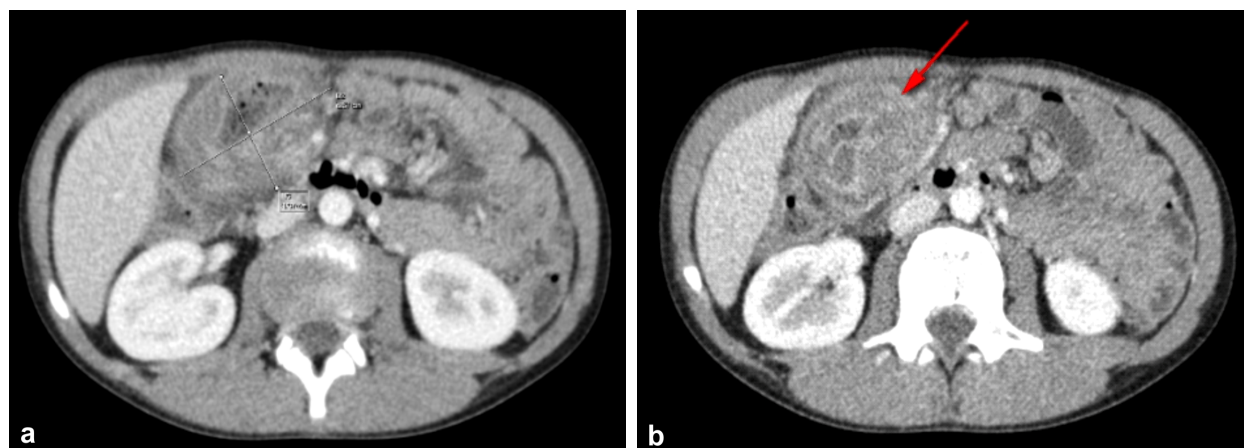


Figure 1 – Axial CT scan showing mesocolic panniculitis: (a) The mesenteric fat is hyperdense compared with the subcutaneous or retroperitoneal fat; (b) “Tumoral pseudo-capsule” differentiating normal mesentery from the inflammatory process.



Figure 2 – Macroscopic appearance of the tumoral mass (impaired mesocolon and hepatic flexure in the colon).

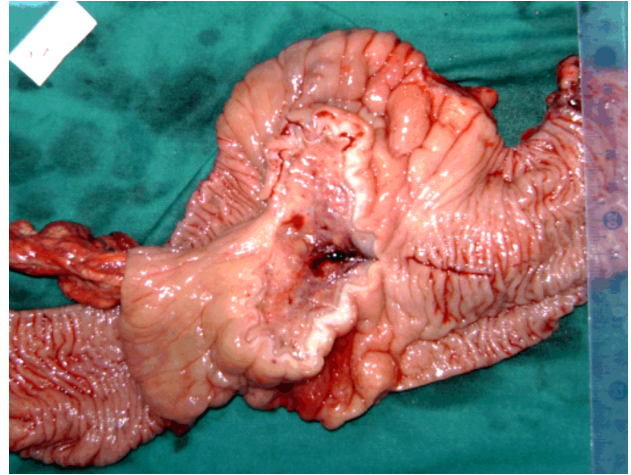


Figure 3 – Macroscopic appearance: longitudinal section (thickened colonic mucosa and appearance of fatty necrosis in the mesentery).

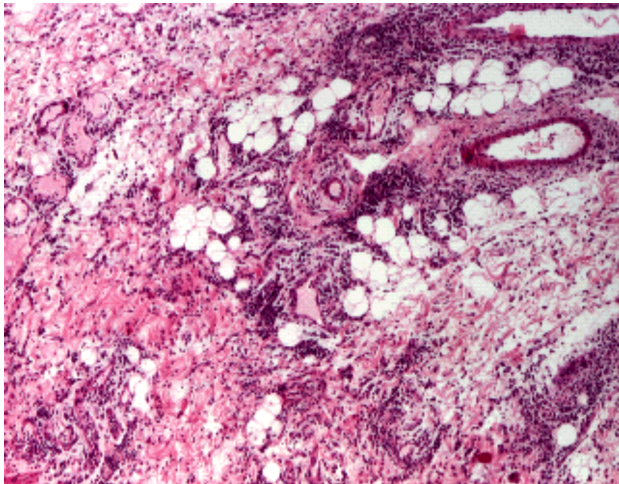


Figure 4 – Mesocolon with fatty infiltration and chronic inflammation [Hematoxylin–Eosin (HE) staining, ×40].

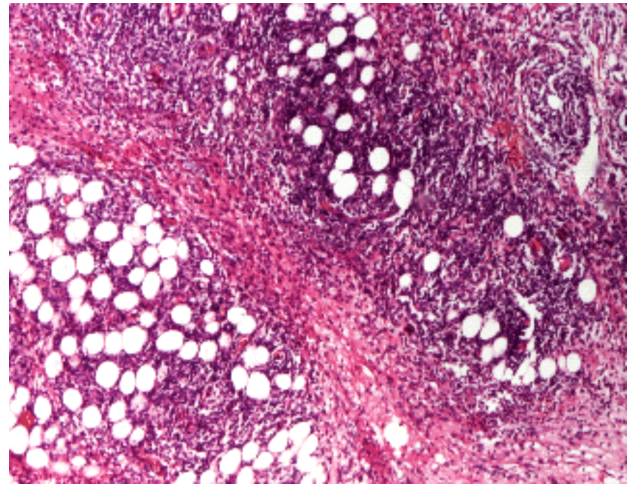


Figure 5 – Mesocolon with non-specific inflammation (HE staining, ×40).

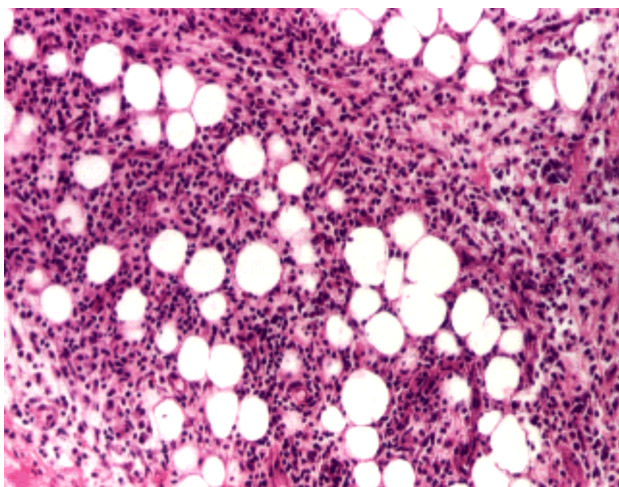


Figure 6 – Polymorph inflammatory infiltrate in the colonic serosa (HE staining, ×100).

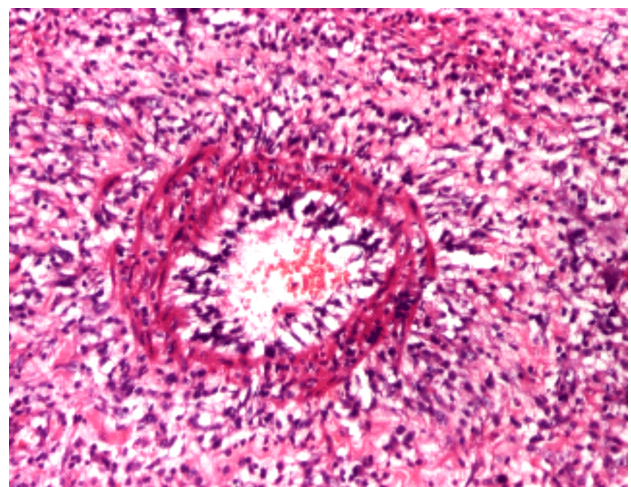


Figure 7 – Obliterant endarteritis at serosal level (HE staining, ×100).

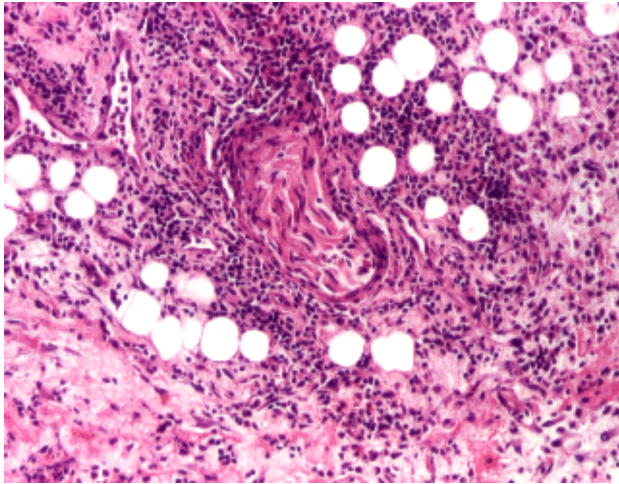


Figure 8 – *Perineuritis at serosal level (HE staining, ×100).*

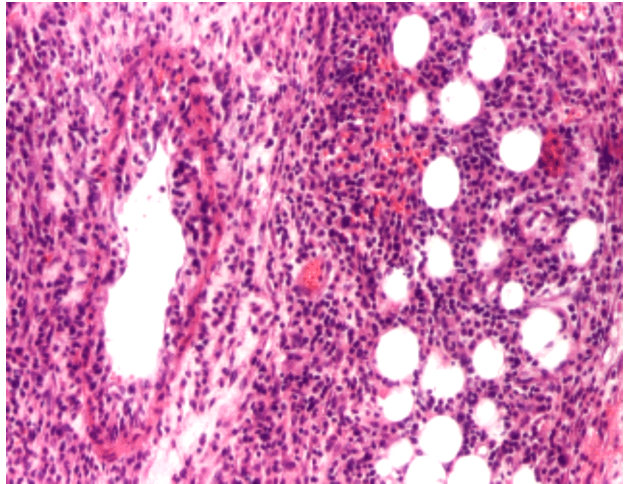


Figure 9 – *Polymorph inflammatory cells in mesocolon with vascular wall impairment (vasculitis) (HE staining, ×100).*

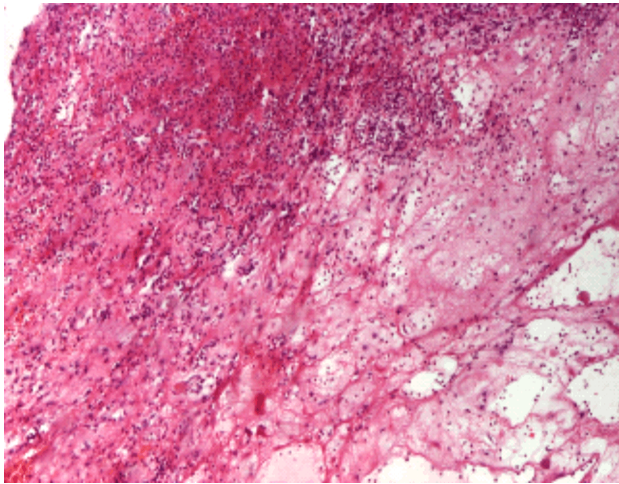


Figure 10 – *Hemorrhagic and edematous areas (HE staining, ×40).*

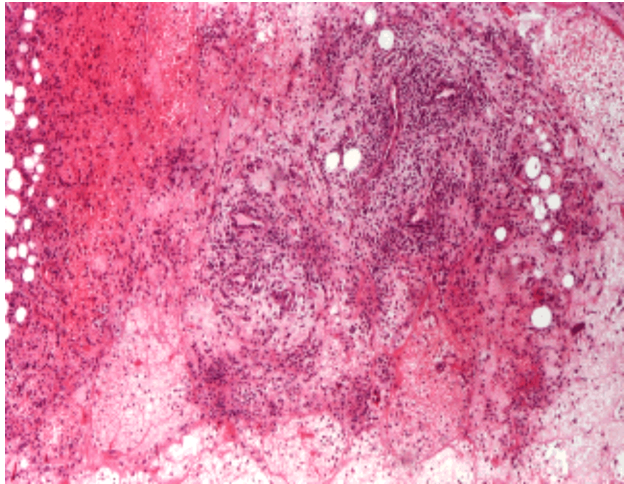


Figure 11 – *Hemorrhage and necrosis in mesocolon (HE staining, ×40).*

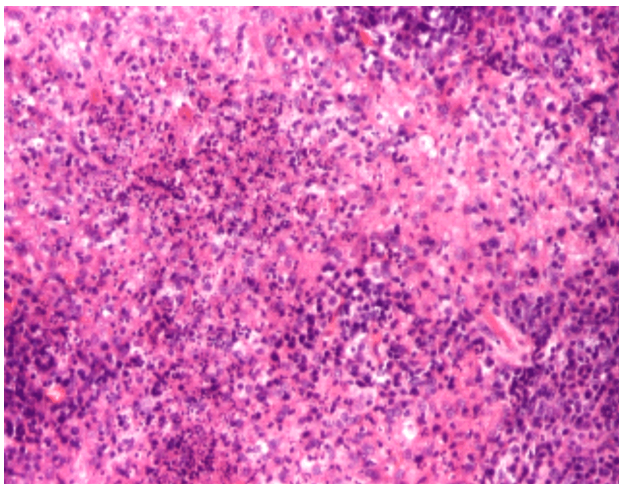


Figure 12 – *Foci of necrosis with dystrophic calcifications (HE staining, ×100).*

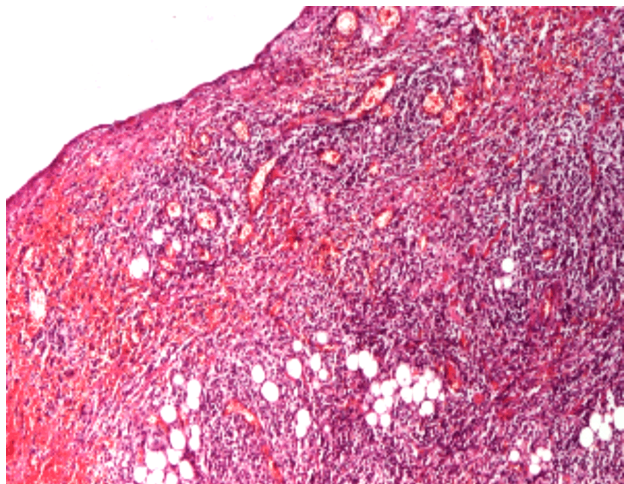


Figure 13 – *Granulation tissue (HE staining, ×40).*

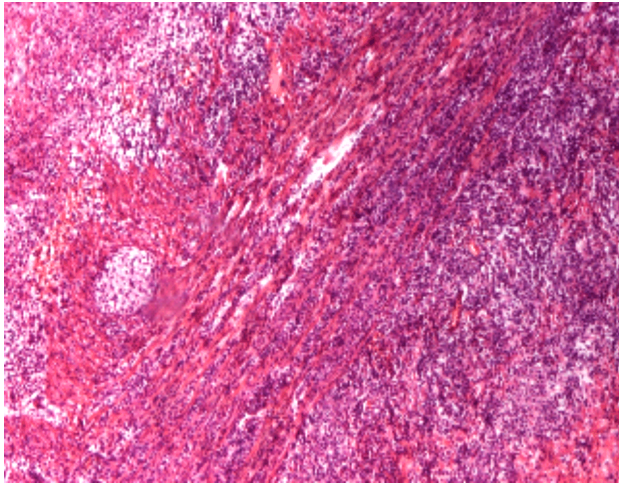


Figure 14 – Massive polymorphous inflammatory cells in muscle layer, affecting the myenteric plexuses (HE staining, $\times 40$).

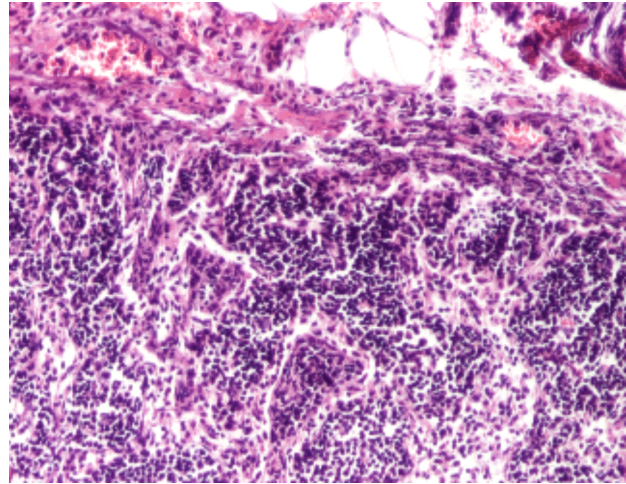


Figure 15 – Lymph node in the mesocolon, with sinusal histiocytosis (HE staining, $\times 100$).

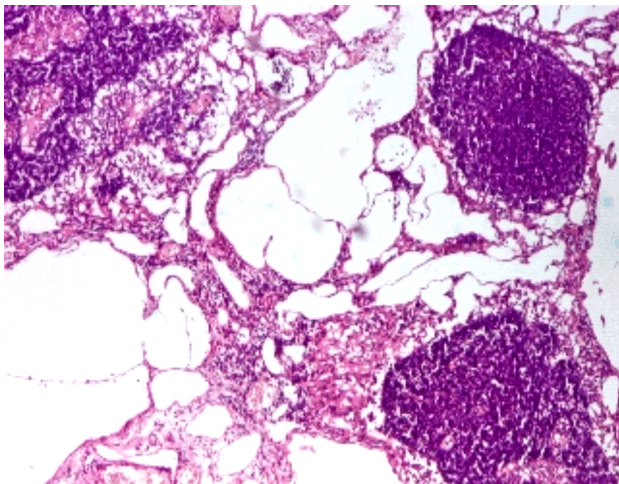


Figure 16 – Lymph node in the mesocolon, with sinusal histiocytosis and dilated lymphatic vessels around the lesion (HE staining, $\times 40$).

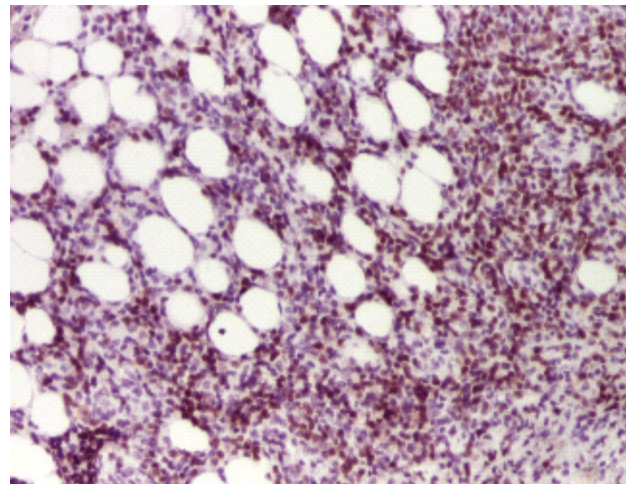


Figure 17 – CD3 is positive in T-lymphocytes (IHC staining for CD3, $\times 100$). IHC: Immunohistochemistry.

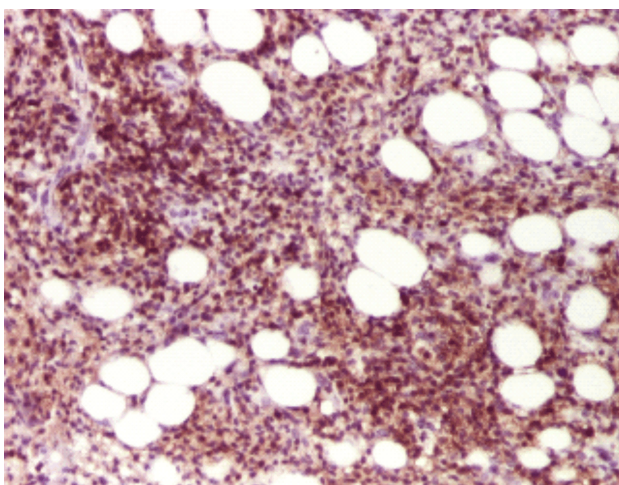


Figure 18 – CD20 is positive in B-lymphocytes (IHC staining for CD20, $\times 100$).

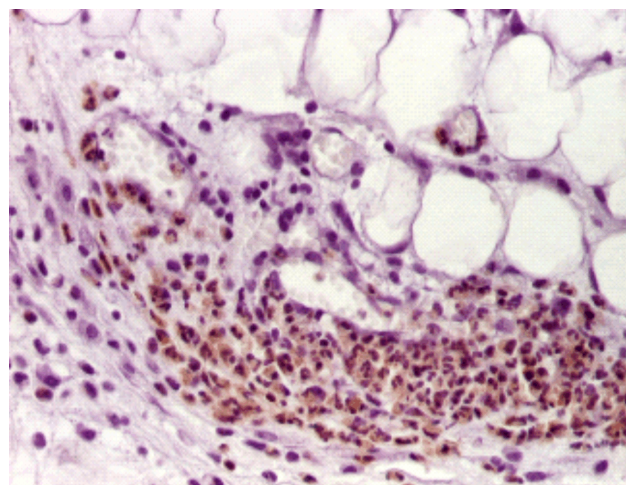


Figure 19 – Leukocyte myeloperoxidase (MPO) is positive in granulocytes (IHC staining for MPO, $\times 200$).

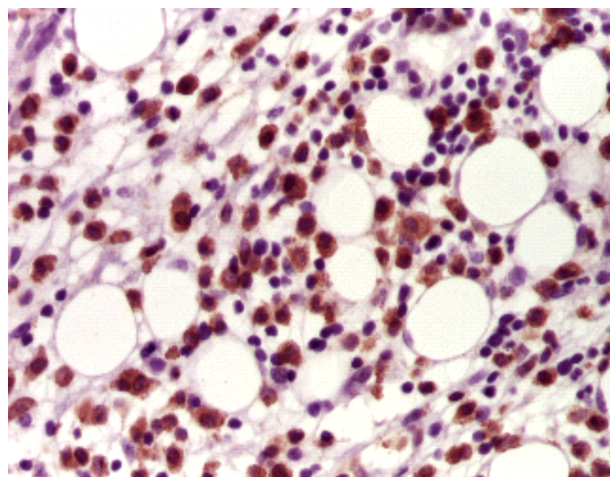


Figure 20 – CD68 is positive in macrophages (IHC staining for CD68, $\times 200$).

Therefore, the transparietal colitis lesions were not specific; the mucosa's glandular architecture was preserved, without basal plasmacytosis, with frequent cryptitis and cryptic microabscesses. The wall had areas with suppurating appearance, perineuritis, acute vasculitis, reactive lymphoid nodes, with marked sinusoidal dilation; no granulomas were detected. The IHC markers did not reveal tumoral aspects lymphoid-like or parietal alterations suggestive of a chronic inflammatory intestinal disease Crohn's-like. The microscopic examination of the thickened mesentery revealed suggestive elements of mesenteric panniculitis, in different proportions: fatty mesentery infiltration, chronic inflammation and less fibrosis; this examination also revealed multiple macrophages with foamy cytoplasm, monocytes or lymphocytes infiltration, necrosis, etc.

Discussion

Mesenteric panniculitis is a rare condition in children, and its location in the colon is also infrequent. Only 17 cases of mesenteric panniculitis were reported in children in literature, in 2010, with an average patient age of 6.5 years and an approximately equal gender ratio. Its etiology is unknown, yet some of the incriminated factors are abdominal injuries, cold exposure, ischemia or intestinal infections. It was found in association with autoimmune or granulomatous diseases, vasculitis, malignancies, pancreatitis or tuberculosis [12, 13]. Issa & Baydoun reported an 84% association rate of mesenteric panniculitis with abdominal injuries or with a history of abdominal surgical procedures, although other studies did not confirm such a strong relation [4, 14]. However, in our patient, a causality relation with the small yet frequent injuries he suffered at the abdominal level during his sports practice twice a week may be hypothesized, although this correlation cannot be proven.

The most frequent symptom on the patients' initial examination was abdominal pain, and some patients even required surgical procedures for acute surgical abdomen, all the more so as the differential diagnosis was difficult, as it included neoplasms, such as lymphomas, carcinoid tumors, liposarcoma, desmoids tumor, retroperitoneal fibrosclerosis, just as in our case report [2, 15]. The disease

may be asymptomatic in up to 30–50% of the cases or it may be accompanied by abdominal pain, vomiting, diarrhea or constipation, weight loss, abdominal tumoral mass detectable on palpation [15]; in 20% of the cases, it is associated with a debilitating chronic evolution or severe morbidity, for instance intestinal occlusion, perforation and peritonitis, and literature even reports a few cases with fatal outcome [16, 17]. Jaundice, rectal bleeding, gastric obstruction and acute abdomen are rarer symptoms [8, 18]. Our pediatric patient came to the hospital with acute digestive symptoms that the conducted paraclinical tests connected to a tumoral mass detected in the hepatic flexure of the colon. Laboratory test values are usually within the normal range in such patients, yet the inflammatory values are sometimes high, just as in our case. The diagnosis may be suggested by an ultrasound scan and especially by a CT scan, revealing fat density increase in the mesentery and a “misty mesentery” appearance (about 40–60 HU retroperitoneal and subcutaneous fat density as compared to the normal values of 100–160 HU). These elements and the almost specific “fat ring sign” were also detected in the reported case. Other elements that may be detected are an encapsulated heterogeneous tumoral mass in over 50% of the cases, located at the root of the mesentery or next to the intestinal loops, the walls of which are thickened yet not actually invaded, mesenteric ganglions smaller than 5 mm (just as in the reported case), condition onset especially in the left side [1, 4, 7, 19]. The mesenteric adenopathies with diameters larger than 10 mm are not suggestive of mesenteric panniculitis and they would require a biopsy in order to rule out any malignancy. The “misty mesentery” appearance is not specific either, as any other infiltrating process in the mesentery needs to be ruled out (edema, hemorrhage, lymphoma, lymphosarcoma, desmoids tumors) [20]. A study conducted on 33 patients selected from literature showed that the diagnosis was set according to a CT scan in 12 patients, according to the biopsy findings after laparotomy in 11 patients and according to the histopathological examination of the resection part (mesentery +/- impaired intestine) in 10 patients [21]. Concerning the therapeutic attitude, 21 out of the 33 cases were administered medication, 12 of whom received steroids (just one case was resistant to this medication), whereas the other 12 patients underwent surgery. In four of the patients, the surgical procedure was chosen to rule out any suspicion of malignancy, since the mesenteritis had a tumor-like appearance and incorporated the afferent bowel section, just like in the patient whose case is reported here. Three other patients were operated on, due to their ileus, three due to their resistance to conservative treatment and two due to their acute pain symptoms. The afferent bowel segment had to be removed in nine of the 12 patients who underwent the operation. One of the patients, in whom bowel resection was not possible, did not recover and died after the operation and two others died due to their resistance to conservative treatment [21]. Another paper reports the case of a five-year-old girl hospitalized for colicky abdominal pains, who was administered conservative treatment at first, but was then operated on and had an appendectomy performed. The abdominal pain persisted after the

operation, the patient's general condition worsened, she was then hospitalized again, examined and given conservative treatment, which resulted into partial symptom improvement. On her fourth hospitalization, she underwent surgery again for the resection of the tumor existing in the ascending colon, which on histopathological examination proved to be mesenteric panniculitis. The patient subsequently recovered [22]. Literature also reports the case of a six-year-old girl with signs of intestinal occlusion, for which surgery was performed and several adhesions were lysed. Although initially positive, the patient subsequently evolved towards many subocclusion episodes (for which she received conservative treatment) with iron deficiency anemia and considerable weight loss. Four months later, a diagnosis of idiopathic sclerosing mesenteritis was established in that case [2]. Therefore, we support an interventionist (surgical) approach to any abdominal tumoral mass detected in children by means of ultrasound or CT scan, especially in case of acute symptoms, as was the case with our patient. Thus, the diagnosis may be set further to histopathological examination and the patient may recover brilliantly.

A positive diagnosis of mesenteric panniculitis may only be set by histopathological methods. The tumor has a nodular macroscopic appearance, just like a lymphoma, and may take on three different forms: mesentery infiltration and diffuse thickening in 42% of the cases, solitary mesenteric mass in 32% of the cases and several tumoral masses in the rest of the cases [12]. Microscopically speaking, there are three suggestive elements found in different proportions, namely fatty mesentery infiltration, chronic inflammation and fibrosis.

Multiple adipose cells with foamy cytoplasm, monocytic or lymphocytic infiltration, lipid- and giant cell-loaded macrophages, necrosis appearance and calcifications may also be detected [23]. Predominant inflammation is suggestive of mesenteric panniculitis, whereas fatty necrosis predominance supports a mesenteric lipodystrophy diagnosis. When fibrotic lesions predominate, the disease is called retractile or sclerosing mesenteritis, the later being considered the final, aggressive stage of the disease [4, 24]. This condition has different names in literature, which actually reflect the histological variety of the reported cases: mesenteric liposclerosis, mesenteric manifestation of the Weber-Christian disease, xantho-granulomatous mesenteritis, inflammatory pseudotumor, mesenteric lipogranuloma, mesenteric lipomatosis and nodular systemic panniculitis, respectively [22]. Since no consensus was reached on its precise nomenclature, it is not sure whether these are separate entities or stages of the same condition. Nevertheless, the names mesenteric panniculitis and sclerosing mesenteritis are the most common. The onset of the inflammatory process is blamed on adipocytokines, *i.e.*, adiponectin, resistin, leptin, interleukin (IL)-6 and tumor necrosis factor- α (TNF- α). The macrophages detected in the adipose tissue are thought to be transdifferentiated from local adipocytes, which supports the assumption that macrophages and adipocytes may be interconvertible, thus involving the mesenteric adipose tissue in conditions such as sclerosing mesenteritis [25].

The treatment of this condition is not standardized, and a conservative approach to the disease is possible, provided it is accurately diagnosed. Thus, corticosteroids, colchicine, immunosuppressants, progesterone, tamoxifen or thalidomide may be administered with good results [2, 8, 26]. When the differential diagnosis with surgical conditions is not positive, the symptoms persist or worsen under conservative treatment or the patient complains from the very beginning of acute surgical abdomen, an aggressive therapy and tumoral mass resection are recommended. Therefore, the prognosis seems to be good, as spontaneous regression may occur after five months in some cases, whereas in other cases the symptoms disappear under conservative treatment. Patients with occlusive or ischemic symptoms require surgical procedures. Nevertheless, chronic inflammatory diseases are associated with increased morbidity in children and, considering that two deaths were reported in children suffering from mesenteric panniculitis in literature [2], all such cases should be made available to the public so as to make this condition known. A recent study conducted in adults has proven a significant association between mesenteric panniculitis and malignancies in general and prostatic carcinoma in particular, as well as a predisposition of these patients to subsequent neoplasms [7]. Yet, these findings have not yet been proven in children due to the small number of reported cases and to the absence of prospective studies.

Conclusions

Although it is an extremely infrequent condition in children and just as infrequent in the colon, mesenteric panniculitis should be known and taken into consideration when setting the differential diagnosis of a chronic or acute onset abdominal painful pathology, especially when the medical imaging tests reveal an abdominal tumoral mass. Once the positive diagnosis is established, the condition may have a positive evolution under conservative or surgical treatment, yet the patient will require long-term follow-up since there is a risk of subsequent association or occurrence of neoplasms.

Conflict of interests

Nothing to declare.

Patient consent

Written informed consent was obtained from the patient's parents for the publication of this report and accompanying images. Ethics Committee approval of the Hospital was obtained for the publication of this article.

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