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Retroperitoneal and iliopsoas abscess as Crohn's disease onset mimicking a common lumbosciatic pain

Crohn's Disease (CD) refers to a chronic transmural bowel inflammation affecting a range between 5 and 15 per 100,000 person-years worldwide [1]. In patients with stricturing CD, the transmural pattern of inflammation may trigger the development of sinus tracts able to connect to other tissues, leading to the formation of fistulas or abscesses [2]. Intra-psoas abscesses are rare, affecting between 0.4-4.3% of patients with CD [3].
We present the case of a 36-year-old male with no past medical history except for a two-week worsening lumbosciatalgia not responding to standard nonsteroidal anti-inflammatory drugs, who complained sudden abdominal pain, with right lower abdominal stiffness combined to a severe edema and erythema of the right lower limb, extended from the gluteus down to the knee, involving the anteromedial and posteromedial areas of his thigh. Patient was septic and CT scans revealed a large complex air-fluid collection within the right iliac region, involving terminal ileum, right retroperitoneum and right lateral abdominal wall towards the inner edge of his thigh.
Diagnosis of CD was made on histopathology and the patient gained full recovery thanks to a prompt surgical intervention followed by high-dose antibiotic infusion and vacuum-assisted wound closure.
Intra-psoas abscesses, albeit rare, are a known manifestation of CD and frequently lead to misdiagnosis because of their rarity and their unusual location easily mimicking other diseases.
Therefore, clinician's awareness must be heightened for complicated CD in the setting of intra-psoas abscesses in order to avoid delayed treatment.

KEY WORDS: Crohn disease, Psoas abscess, Sciatica, Late onset disorders, Negative pressure Wound therapy

Introduction

CD is a multifactorial chronic inflammation that can affect any part of the gastrointestinal tract, with the terminal ileum being the most frequent location. Few retrospective studies have shown a 10% of patients developing spontaneous intra-abdominal abscess within the first 5 years after the diagnosis. According to the Montreal Classification, CD phenotype may be divided into three categories: stricturing, penetrating and non-stricturing/non-penetrating [4]. The full thickness inflammation of the intestinal wall may lead to strictures, fistulas and abdominal abscesses [5]; fistulas can establish enteroenteric, enterocutaneous, enterovesical and enterovaginal connections or even extend posteriorly through the retroperitoneum and iliopsoas space. The mechanisms underlying the formation of these abscesses include transmural bowel inflammation with fistulization and direct penetration of bacteria from the diseased bowel to con-

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tiguous tissues, remote blood seeding of bacteria from the diseased bowel, and peritoneal contamination during surgery [6]. Spontaneous abscesses are most commonly located in proximity to the ileocecal area. Surgical pathological examination of resected bowel specimens has demonstrated the association between penetrating disease and mechanical factors, such as luminal narrowing of the bowel from fibrotic stricture, explaining why the presence of strictures and fistulas is frequently associated with the failure of nonsurgical approach and the necessity of surgery with bowel resection [4]. At least 80% of abscesses contain multiple bacterial types, often a mixture of aerobic and anaerobic flora, especially Escherichia coli, Enterococcus fragilis and Peptostreptococcus species. Abscesses may be seen as the result of a complex interaction between the bacteria and the host, enhancing the accumulation of neutrophilis in the infection along with the deposition of fibrin which encases the area and entraps the bacteria together with inflammatory cells. A number of serologic and genetic markers have been associated with the development of intra-abdominal abscess in patient with CD. As a matter of fact, anti-Saccharomyces cerevisiae antibody (ASCA) has been strongly associated to penetrating and fibrostenosing CD. ASCA immunoglobuline A or G positive patients appear to have an increased risk for requiring surgery in the early stages of the disease. Other serologic markers including Escherichia coli outer membrane-porin (OmpC), CBir1 flagellin, antilaminaribioside carbohydrate (Alca) and antichitobioside carbohydrate (Acca) have been linked to internal-penetrating CD. However, the clinical value of using these markers for clinical purposes has not been pursued [6].

Symptoms of abdominal abscesses can be unspecific and most commonly reveal themselves as severe pain, fever, diarrhea and nausea; presence of acute peritonitis or free perforation from CD are both extremely rare. Mostly, spontaneous intra-abdominal abscesses are located in the right lower quadrant involving the pelvis or the proximity of psoas muscle with flank or back pain and claudication. As known, intra-abdominal abscesses are a challenging occurrence in surgical practice due to a wide range of differential diagnoses and overlap of nonspecific symptoms. In our experience we assisted at the extremely rare onset of a severely complicated CD mimicking a two-week worsening lumbosciatic pain.

The differential diagnosis of sudden onset of claudication and pain in the hip in a patient with intra-abdominal and retroperitoneal abscesses radiating to the inferior limb should include the presence of an undiagnosed CD.

Case report

A 36-year-old male, with no past medical history except for a two-week worsening lumbosciatalgia, was firstly evaluated from his general practitioner who scheduled a spine MRI showing intramedullary spinal cord abnormalities associated to an enlarged right iliopsoas muscle believed to be a simple hematoma. Thus, the patient was referred to an orthopedic surgeon who prescribed standard nonsteroidal anti-inflammatory drugs with no significant relief. A few days later the patient was transferred to our department from the emergency room of a smaller Hospital in a severe septic condition. On physical examination, the patient presented with tenderness in the right lower abdomen and right hip combined to a severe edema and erythema of the right lower limb, extended from the gluteus down to the knee involving the anteromedial and posteromedial areas of his thigh (Fig. 1). Vital signs upon admission showed T>39°C, severe tachycardia, hypotension and nausea. Admission lab results revealed an increase of WBC mostly neutrophil (18.000 WBC, N= 94%), platelet count of 640.000/μL, hemoglobin 11.3 g/dL (reference range 13-17), RCP 39 mg/dL (reference range 0.0-0.5), PCT 9.03 ng/mL, while COVID19 PCR test turned out negative. Contrast-enhanced CT revealed a large complex air-fluid collection within the right iliac region, involving terminal ileum, right retroperitoneum and right lateral abdominal wall towards the inner edge of his thigh (Fig. 2A). The patient underwent urgent surgery. On laparotomy, vermiform appendix appeared to be gangrenous, tightly adhered to the cecum and the small intestine through multiple bridles and adherences as a result of a likely complicated pelvic and retroperitoneal abscess, sec-

Fig. 1: Upon admission, physical examination showed severe edema and erythema of the right lower limb, extended from the gluteus down to the knee involving the anteromedial and posteromedial areas of the right thigh.

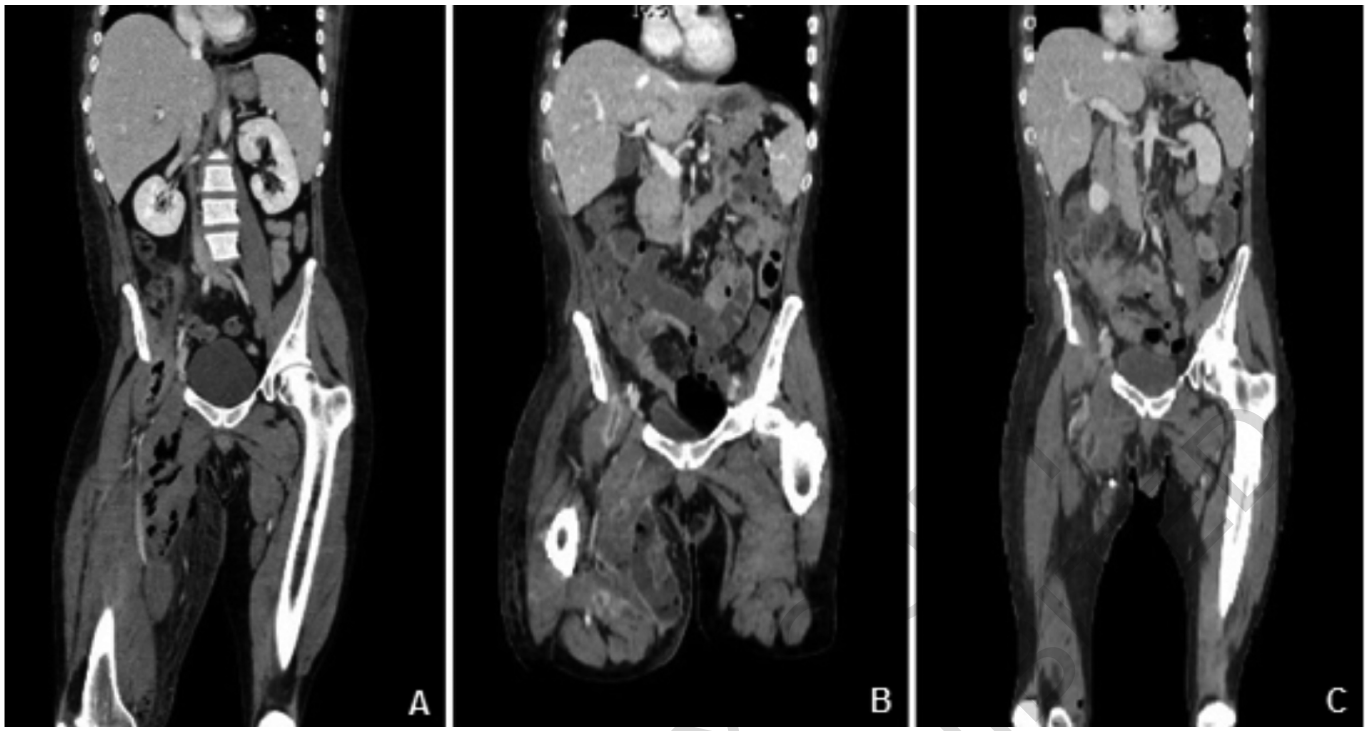

Fig. 2: A) CT scan upon admission revealed a large complex air-fluid collection within the right iliac region, involving terminal ileum, right retroperitoneum and right lateral abdominal wall towards the inner edge of the right thigh; B) CT scan, seventeen days after surgery, showed few residual air-fluid collections within the inner edge of the patient's right thigh, involving iliacus, pectineus, adductor and sartorius muscles towards both the subcutaneous and cutaneous tissues anteriorly. Therefore, the radiologist using the imaging guidance placed a 10 French percutaneous abscess drainage in the biggest and declivous collection of the right thigh; C) One week later a new CT scan showed a significant downsizing of the right inferior limb collection and the drainage was soon after removed.

ondary to fistulous connection between the cecum, terminal ileum and right iliopsoas muscle, spilling pus all around (Fig. 3). Therefore, abdominopelvic peritoneal washing and uneventful ileocecocolic resection were performed, followed by a hand-sewn ileo-colic latero-lateral anastomosis. Vascular surgeon was called and a right longitudinal inguinotomy followed by drainage and debridement of the necrotizing soft-tissue infection along the sartorius and the adductor muscles were performed. Both a peritoneal drainage and a subcutaneous drainage in his right thigh were placed. Intraoperative cultures taken from the abscess eventually grew Escherichia coli, Enterococcus faecium and Streptococcus constellatus. The patient was started on high dose of intravenous piperacillin-tazobactam, tigecycline and fluconazole with appropriate response.

On fifth postoperative day, the patient underwent lower extremity venous Doppler evaluation. Despite the compromised status of the inferior right limb, Doppler measurements obtained from bilateral common and superficial femoral, bilateral great saphenous, and bilateral popliteal veins, did not show any noteworthy finding. He was given heparin as a four-week post-operative prophylaxis.

Besides regular bedside wound care, four days after surgery, the patient returned to the operating room, where his wounds were abundantly washed, disinfected and packed with iodoform gauzes and his inferior right limb drainage was removed. A significant reduction of pus oozing was observed and four days later a vacuum-assisted wound closure device was successfully placed on his thigh (Fig. 4). Seventeen days after surgery, an abdominal and right inferior limb CT was scheduled. Few residual air-fluid collections were revealed within the inner edge of the patient's right thigh, involving iliacus, pectineus, adductor and sartorius muscles towards both the subcutaneous and cutaneous tissues anteriorly (Fig. 2B). Therefore, the radiologist himself placed a 10 French percutaneous abscess drainage using the imaging guidance in order to allow the catheter to reach the biggest and declivous collection of the right thigh. Cultures from the drainage eventually grew Streptococcus constellatus and the antibiotic therapy was confirmed. One week later a new CT showed a significant downsizing of the right inferior limb collection and the drainage was removed (Fig. 2C). Eight days later the vacuum-assisted wound closure device was removed and the patient was discharged fully recovered.

During recovery, Physiotherapy was called for evaluation and the patient was started on a rehabilitation program that helped him fully regain walking ability within a month from surgery.

Fig. 3: Nelaton catheter introduced through the fistulous connection between the retroperitoneum and the iliopsoas right muscle.

Fig. 4: Eight days after surgery, a vacuum-assisted wound closure device was placed on the right thigh, significantly improving wound healing.

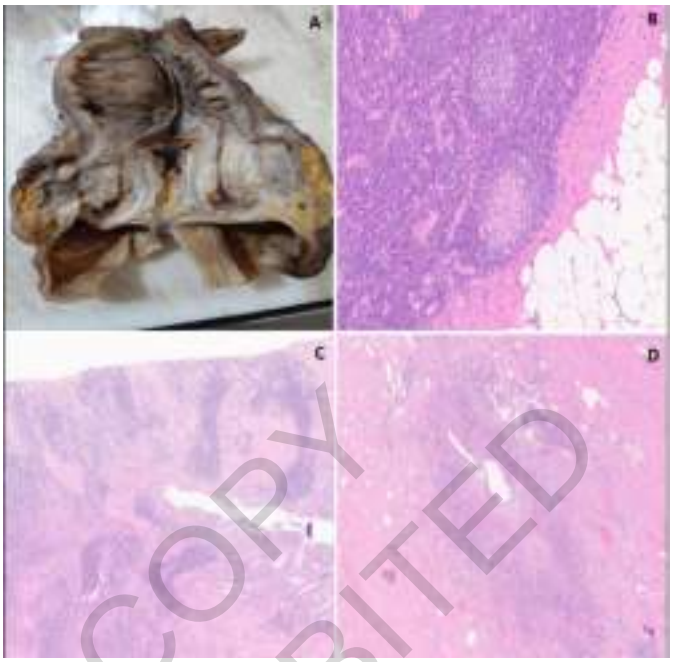

Fig. 5: A) Pathological specimen of the resected cecum and 50 mm of terminal ileum, showing thickening of the pericecal adipose tissue surrounding the vermiform appendix, associated to fibrin deposits on the serosal surfaces; B) Microscopic examination of the surgical specimen revealing a lymph node section containing a lymphoid follicle with a contiguous granulomatous lesion; C) Microscopic examination of stained ileocecal valve tissue section showing mild architectural glandular disarray, erosion of the epithelial lining, numerous follicular lymphoid aggregates within the submucosa and between the muscularis propria and the serosa. D) Microscopic evidence of the appendiceal lumen characterized by de-epithelialization of the mucosa and chronic active inflammation with both lymphocytes and neutrophil granulocytes represented.

Microscopic examination of the surgical specimen revealed circumscribed inflammation, basal plasmacytosis and, mostly, transmural lymphoid aggregates compatible with ileocolic CD, extended to the vermiform appendix and the ileal resection margin (Fig. 5).

Thus, the histologic examination of the resected specimen has been the key step for the diagnosis and differential diagnosis, particularly in the differentiation of ileocolic CD and other non-IBD related pelvic and retroperitoneal abscesses.

Giving those findings, the patient was referred to Gastroenterology for follow-up and any future medical treatment.

Both early outpatient follow-up and three-month outcome demonstrated excellent recovery.

Discussion

CD refers to a chronic transmural bowel inflammation affecting a range between 5 and 15 per 100,000 person-

years worldwide [1]. Although it is regarded as being a common disorder, it has not failed to arouse researcher's and practicing physician's attention on account of its extremely variable clinical features. It may involve every part of the gastrointestinal tract, but in the majority of cases it mainly affects the terminal ileum and/or the colon. The disease is characterized by the presence of extraintestinal manifestations and perianal complications in about 30% of patients, as well as the development of intra-abdominal or pelvic abscesses in 10-30%. Fistulas and abscesses may present insidiously, without any apparent orifice, usually localized in the perianal region and treatment is rather easy; in other cases fistulas are more complex and usually resulting from a subclinical penetration of the intestinal wall. Once fistulas connect to a pelvic or retroperitoneal abscess, they may extend along another anatomical region, tracking through the obturator foramen or along the iliopsoas muscle into the hip and thigh. In such cases, the clinical symptoms may be misleading or mild and the diagnosis may be wrong or delayed. Therefore, the diagnosis of a fistula or an abscess secondary to an undiagnosed CD should always be born in mind, even when affecting atypical sites. Mayer et al. reported that a spontaneous retroperitoneal abscess in a patient with CD does not always terminate at the psoas or iliac spaces but it can pass through the sciatica foramen and result in a buttock or posterior upper thigh abscess. Otherwise, if passing through the obturator foramen, the abscess may spread to the thigh anteriorly. Psoas abscesses may be a complication of CD as well as appendicitis, diverticulitis or metastatic colorectal cancer, while in the past tuberculosis of the spine, among the infectious etiologies, was the commonest cause [7]. They are referred to as an uncommon disease entity causing considerable morbidity and mortality, classified as primary or secondary, depending on the presence of underlying disease. Primary cases commonly occur in association with diabetes mellitus, intravenous drug use, AIDS, renal failure and immunosuppression, while secondary causes are usually due to infective or inflammatory conditions affecting intra-abdominal structures. Psoas muscle lies in proximity to many structures, including jejunum, ileum, colon, vermiform appendix, aorta, renal tract, pancreas and spine; thus infection of any of these anatomical structures can spread and involve the adjacent psoas muscle. It is thought that the rich vascular supply to the muscle predisposes it to haematogenous spread from sites of occult infection [8]. Diagnosis is made after accurate evaluation of patient's medical history, careful physical examination, laboratory assessment and imaging studies. CT and MRI are considered the most sensitive and specific imaging tests, whereas US, especially in detecting deep pelvic or retroperitoneal abscesses, besides being operator dependent, has shown a mean sensitivity and specificity of 91.5% and 93% respectively. Radiologic imaging is important not only for the detection of abscesses, but also for the identification of any associated fistula, a finding which can alter patient management [6]. Maconi et al reported transabdominal bowel sonography as very useful in the early diagnosis and surgical planning of CD complications, such as stenosis, phlegmons, abscesses and fistulas, adding that Endoscopic UltraSonography has a wide use in the follow-up of operated patients, detecting endoscopic recurrence, especially in case of extended resections without free margins [9]. Due to the psoas muscle innervation from L2, L3, and L4 nerves, symptoms that should arouse suspicion of an iliopsoas and retroperitoneal abscess in patients with CD are hip pain, difficulty in walking, pain in the buttock or thigh, fever and profundus iliac fossa tenderness. Flexion contracture, extremity tenderness, a limping gait and pain in the limbs appear to be more specific symptoms, especially in those patients known to have CD [10]. Ricci M. et al. described thromboembolism as a possible complication, rarely causing paresis of the lower extremities due to its direct spread through the hip with consequent septic arthritis or through the spinal cord resulting in extradural spinal abscess [11]. As a matter of fact, in our experience, the patient underwent lower extremity venous Doppler evaluation not showing noteworthy findings, however he was given heparin as post-operative prophylaxis recommends. RCP and ESR are almost invariably elevated as well as leucocytosis with left shift. Although the advantages in medical treatment, often combined with percutaneous drainage, have reduced the indication to a surgical intervention, surgery remains mandatory especially when initial medical treatment fails or obstructions, toxic megacolon, perforations with diffuse peritonitis, abscesses, fistulas or bleeding occur. One of the possible causes of sepsis and of the unfavorable outcome is the bacterial translocation [5]. The European Crohn's and Colitis Organization (ECCO) recommends that a CD patient with a spontaneous intra-abdominal abscess should be managed with broad-spectrum antibiotics, imaging-guided percutaneous drainage or surgical drainage followed by delayed resection, unless patient's critical clinical conditions urge an immediate surgery [12]. Medical management of these infections is aimed to treat mixed aerobic and anaerobic bacterial infections. According to scientific works published by Rogueiro et al. and Yoshida et al, there is strong evidence that starting TNF- inhibitor therapy within 6 months after an ileocecal resection prevents postoperative recurrence [13-14]. Colombo et al. underline that, according to Vienna Classification – a simple phenotypic classification structured on a combination of age at diagnosis, location and behavior of disease, in order to categorize Crohn's patients into various subgroups – penetrating behavior (B3) is confirmed to be an important risk for early postoperative recurrence; therefore, patients with penetrating B3 disease (i.e. fistulas) have a specific indication for immunosuppressive or anti-tumor necrosis factor-α–therapy [15]. As for placing percutaneous drainages, which is

believed to be a relatively safe procedure, their success rate strongly depends on abscess's size. It has been suggested that abscesses >4 cm in diameter will not resolve without drainage, while those ≤4 cm are likely to respond to antibiotics. The American College of Radiology recommends imaging-guided percutaneous drainage for CD-related abdominal fluid collections greater than 4 cm. Reported efficacy with percutaneous drainage in CD abdominal abscess ranged from 50% to 92%. Sometimes the small-caliber catheters may be easily obstructed by necrotic tissue, blood clots and thick purulent fluid in the abscess cavity. In those cases, greater caliber catheters could replace the small-sized drainage system. Liu et al. have described a strategy using a sump drain with trocar puncture and continuous irrigation and suction under negative pressure, in order to avoid blockage of the drainage system and allow for faster resolution of abscess [4]. As demonstrated in our experience, in severe septic cases, transabdominal resection of the diseased bowel, retroperitoneal debridement and external drainage of the abscess cavity are necessary. Most importantly, the resection of the involved bowel allowed us to finally identify the patient's condition with CD in order to refer him to a proper medical follow-up. In our case, vacuum-assisted closure represented an alternative and viable method of wound management; thanks to negative pressure and a thorough debridement, adequate haemostasis and application of sterile foam dressing, vacuum-assisted closure stabilizes the wound environment, reduces wound edema and bacterial load, improves tissue perfusion and angiogenesis, giving the chance of achieving primary closure of wounds and significantly reducing the need for plastic procedures. Although surgery may treat the acute septic complication of CD, it is important to recognize that surgery usually doesn't cure CD, as recurrence can be found at the anastomosis in 73% to 93% of cases and clinical recurrence is seen in 20% to 30%. However, prospective studies to delineate factors to determine which patient would be best treated with medical therapy alone, medical therapy plus percutaneous drainage, or surgery are sorely needed [6]. The best strategy of treatment is controversial because the current literature is based on retrospective series.

Conclusions

Psoas abscess is a well-documented complication of CD, yet it rarely presents as the onset of the disease, with only a handful of cases reported in literature [8]. It usually has an insidious clinical presentation, with non-specific symptoms and signs, leading to delays in diagnosis and considerable morbidity and mortality. Notably, to the best of our knowledge, clinician's attention should be paid to the possibility of diagnosing an unknown CD in order to start a prompt wide ranging and multidisciplinary treatment.

The differential diagnosis of sudden onset of claudication and pain in the hip in a patient with intra-abdominal and retroperitoneal abscesses radiating to the inferior limb should include the presence of an undiagnosed CD.

The treatment of major complications of Crohn's disease remains a challenge.

It is felt that radiological-guided drainage is effective for primary psoas abscesses, but secondary abscesses generally require a more definitive surgical procedure, aiming to drain the abscess and treat the underlying cause.

It is important to emphasize that current literature is based on retrospective studies with bias, issue of sample sizes, and some contradictory results. The definition of treatment success is heterogeneous and some studies suffer from a short follow-up. Prospective and larger studies are necessary in order to better understand the impact of CD spontaneous sepsis in the healthcare system and to choose the best treatment options in different clinical scenarios [4].

Riassunto

Il Morbo di Crohn consiste in un'infiammazione cronica intestinale ad eziologia multifattoriale in grado di colpire qualsiasi parte del tratto gastrointestinale con un'ampia variabilità clinica. In pazienti con forme stenosanti della patologia, il caratteristico pattern transmurale dell'infiammazione può indurre la formazione di tragitti fistolosi o di ascessi con il conseguente interessamento delle strutture limitrofe. La concomitante presenza di ascessi retroperitoneali e del muscolo ileopsoas rappresenta una complicanza rara, caratterizzata da un ampio ventaglio di diagnosi differenziali e spesso mascherata da sintomi aspecifici correlabili a disturbi differenti. Ancor più rara è la presentazione di un simile quadro, severamente esteso all'arto inferiore omolaterale, quale esordio di Morbo di Crohn. Nella nostra esperienza riportiamo il caso di un paziente di sesso maschile di 36 anni affetto da persistente dolore lombare destro irradiato all'arto inferiore e associato ad evidenza RMN di presunto ematoma del muscolo ileopsoas destro, interpretato dallo specialista ortopedico quale lombosciatalgia non rivelatasi in seguito responsiva alla terapia antinfiammatoria prescritta. Il paziente giungeva alla nostra osservazione in grave stato settico, quale trasferimento urgente da pronto soccorso di altro presidio. L'intervento chirurgico urgente ha messo in evidenza in fossa iliaca destra un conglomerato di anse intestinali, tenacemente adese alla doccia parieto-colica destra con presenza di tramite fistoloso tra l'ultima ansa ileale, il ceco ed il muscolo ileopsoas omolaterale dal cui spessore fuoriusciva un'abbondante quantità di materiale purulento; una consensuale inguinotomia longitudinale destra ha permesso l'evacuazione di una vasta raccolta purulenta tra i muscoli sartorio e adduttore dell'arto inferiore destro. Il posizionamento di

drenaggi intraoperatori e successivamente di drenaggio percutaneo TC-guidato, insieme ad alti dosaggi di terapia antibiotica infusiva e a presidi medicali a pressione topica negativa hanno permesso una completa restitutio ad integrum con ripresa da parte del paziente, grazie ad un piano riabilitativo individualizzato, della piena autonomia motoria. L'analisi istopatologica ha permesso la diagnosi di Morbo di Crohn ileocolico ed il successivo invio del paziente allo specialista gastroenterologo per l'inquadramento e l'eventuale trattamento medico della patologia. Il trattamento delle complicanze maggiori del Morbo di Crohn resta una sfida, specialmente se coincidente con il suo esordio clinico. È fondamentale sottolineare come l'attuale letteratura scientifica manchi di studi ampi e prospettici che consentano di meglio comprendere l'impatto della sepsi spontanea Crohn-relata sulla Sanità e di individuare le migliori scelte di trattamento per i diversi scenari clinici di manifestazione.

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