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RESEARCH ARTICLE

EXTRA PELVIC ENDOMETRIOSIS: DOUBLE ILEAL AND ILEO-CAECAL LOCALIZATION REVEALED BY ACUTE SMALL BOWEL OBSTRUCTION, A CLINICAL CASE

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Abstract

Endometriosis is a frequent pathology in women, the double ileo-caecal localization is extremely rare. Given the non-specific nature of the clinical symptoms, the diagnosis of this pathology was initially disconcerting. We report the case of a young patient, presenting chronic abdominal pain.Even though she didseveral medical consultations, the diagnosis was not possible given the non-specific character of the symptomatology. Our patient was admitted to our department for an acute small bowel obstruction.The abdominal CT scan was in favor of an ileo-caecalthickening. An ileo-caecal resection was performed by laparoscopy, thus solving the problem of surgical emergency andachieving complete excision of a double tumor of the terminal ileum and the caecum. The endometriotic nature was revealed by the anatomopathologicalanalysis of the surgical specimen.

Introduction:-

This gynecological pathology can affect the urinary or digestive tracts. Abdominal endometriosis may be localized in the right diaphragmatic dome or the recto uterine pouch. Recto-sigmoidal involvement is very frequent, given its proximityto the uterine cervix. Involvementof the ileocolic region by genital endometriosis is possible but rare. We report the case of a patient operated for anacute bowel obstructionwiththe suspicion of a tumor of the ileum. After anatomopathological analysis of the surgical specimen, the diagnosis of an endometriosis was retained.

Observation:-

The patient was a single 34-year-old women, showing a normal menstrual cycle and with no particular medical history.

She had been suffering from abdominal pain without any specific characteristics for many years. The pain required several medical consultations and ceased after a symptomatic treatment byPPI, analgesics, and antispasmodics.

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She was admitted in emergency for an acute small bowel obstruction that had been evolving for 5 days, manifested by abdominal pain, vomiting and no constipation.

Examination showed a stable patient, with a BMI of 25, apyretic and with normal vital signs, a slightly distended abdomen and presenting a simple epigastric and right iliac fossa tenderness.

The biological tests were normal: hemoglobin level at 12 g/dl, leukocytes at 9800/mm3, platelets at 329000/mm3, CRP was high at 174mg/L, Lipase at 21U/L (normal), her kidney functiontests were also normal with a urea at 0,35g/L, creatinine at 7mg/L, blood glucose levels at 1g (normal). Her electrolyte blood testwas also normal.

Tumor markertests were normal: CEA at 2µg/l, CA19 9 at5 U/ml and AFP was normal.
Plain abdominal X rays show hydro-aeric levels(Figure 1).

The CT scan showedbowel dilatation at 28 mm, and hydro-aeric levels. There was an ileocaecal thickening, irregular, and stenosing,measuring 26 mm, surmounted by a distal small rounded lesion at the ileum. No radiological signs of appendicitis (Figures 2,3,4). There was a subserous uterine fibroid of 30 mm.

The CT aspect was in favor of an ileocaecalthickeningsuggesting a Crohn's disease, or an ileal tuberculosis or tumor (adenocarcinoma, lymphoma).

Colonoscopy reveals a complete stenosis of the ileum which the biopsy was normal (non-specific inflammatory lesions). the patient was operated by laparoscopy, using four trocars (Figure 5). Two smallstenosing tumor lesions were discovered, the first atthe ileo-caecal junction and the second at the ileumnear the ileo-caecal junction(Figure 6).

There were no hepatic metastases, and no peritoneal carcinomatosis. There wereno evident lymph nodes. The shapeof this lesion would suggest a neuroendocrine tumor (carcinoid) due to their small size, the color and stenosing aspect. There were no pelvic lesions except for a small uterinefibroid.

An ileo-caecal resection was performed with a side to side ileo-colic anastomosis. The extraction of the specimen was done through a small Pfannenstiel incision. The postoperative course was normal, and the patient remained asymptomatic during the first year.

The anatomopathological analysiswas in favor of a double localization of an endometriosis on the ileo-caecalregion.

Discussion:-

Endometriosis refers to the presence of endometrial tissue with both glands and stroma outside the uterine cavity [1].

Classification

Three forms of endometriosis are described [2]:
– Superficial peritoneal
– endometriomas (ovarian endometrioma)
– Deeply infiltrating

Adenomyosis is different, and is a special type of endometriosis that develops in the uterine muscle[3].

These lesions are often associated due to the multifocal nature of the disease.

Deep infiltrating endometriosis is described as anterior when it involves the bladder and posterior when it involves the uterosacral ligaments, the rectovaginal septum or the rectosigmoid wall.

The main symptom of deep infiltratingpelvic endometriosis is pelvic pain, whose intensity is not always correlated with the infiltrationdepth.
The incidental discovery of minimal and asymptomatic endometriosis has no known pathological relevance, and in untreated patients, the evolution may be spontaneously favorable.

In the current state, there is no indication to treat uncomplicated and asymptomatic endometriosis, regardless of the stageof the disease [4].

Physiopathology

The abnormal presence of endometrial tissue in the gastrointestinal tract can be explained by [3]:

-The reflux of endometrial issue cells into the abdominal cavity through the fallopian tubes at the time of menstruation

-Or by congenital disorders, Mülleriantissue residues which will give endometrial tissue in an ectopic situation [2].

The abnormal endometrialgrafted tissue is sensitive to the woman's hormones.Microscopic bleeding occurs in the affected organ at the time of menstruation, which causes troublesome symptoms. Outside of the menstrual period, endometriosis nodules, when large, can also cause troublesome symptoms.

Intestinal endometriosis initially takes the form of small superficial serous nodules. The cyclical bleeding of the endometriomas lead to local inflammation with infiltration of the wall by fibrosis which causes adhesions to the neighboring organs and stenosis [5].

Epidemiology

Endometriosis is a gynecologic disorder affecting 5% to 10% of sexually active women, 15% of infertile women, and 30% of infertile women with chronic pelvic pain [2].

Women with endometriosis have a short menstrual cycle, prolonged and heavy menstrual bleeding, and early menarche before the age of 12.

Endometriosis can sometimes appear as early as puberty, but most often it appears in women of childbearing age, with a peak in frequency between the ages of 25 and 35. Exceptionally endometriosis is diagnosed at menopause [6].

Endometriosis is reported in women with a high socio-economic status.In fact, the more financial means one has the earlier one consults. This is found whatever the age of the patient at the time of her first pregnancy, whatever her fertility status, and whatever the time elapsed between menarche and first pregnancy [7].

The absence of pregnancy is associated with a greater prevalence of endometriosis. Finally, there is probably a familial predisposition in women with endometriosis.

Rectosigmoid involvement is found in 6-30% of deep infiltrating endometriosis and in 35% of digestive involvement in case of ovarian endometrioma. [2].

It may be unifocal or multifocal in 15% of cases, often associated with deep pelvic and retrocervical involvement infiltrating one or more neighboring organs. Our patient had an ileal and caecal bifocal form without pelvic involvement apart from a uterine fibroid.

Extra-pelvic localizations of endometriosis are exceptional and represent only 5% of cases of endometriosis[5]. There are extra pelvic gastrointestinal, parietal, pleuropulmonary and diaphragmatic localizations. Extraperitoneal endometriosis (lung, pleura) is explained by "metastatic" diffusion by hematogenous or lymphatic dissemination [5].

Gastrointestinal endometriosis can develop outside the pelvic cavity. In a study of 3,037 cases, Weed reported intestinal involvement in 5.3% of patients[5].

Rectosigmoid involvement represent 72% of gastrointestinal involvement, the ileum is involved in 7%, the caecum in 4%, the appendix in 3%, and the stomach, jejunum, transverse colon, and pancreas in less than 1% of cases [6].

Regardless of the mechanism, ectopic endometrial tissue remains hormone-dependent and subject to variation with the menstrual cycle.

Symptomatology

There are no pathognomonic signs of gastrointestinalinvolvement in endometriosis, but it should be suspected in case of:

- Dysmenorrhea[2],
-Sometimes patients present with dyspareunia [6] or,
- Dyschezia[2].

Dyspareunia:pain during deep penetration may be related to rectal involvement.

These pains may be chronic pelvic pain and not related to the menstrual cycle.

Our patient had chronic pelvic pain and had been to the emergency department several times.

Some non-specific signs may appear, without any obvious explanations and may persist despite treatment of the disease, such asintense fatigue, epigastric pain, back pain, meteorism, constipation or diarrhea.

Painful defecation and rectal bleeding at the time of menstruation are specific to endometriosis of the sigmoid and rectum.

Koenig's syndrome can occur in the case of a small bowelendometriosis,it associates pain and abdominal distension, constipation followed by diarrhea [1].

Some cases of intestinal intussusception or peritonitis on perforation of the small bowel have been reported in the literature [8]. The terminal ileum is most often affected, generally it is formed by a single nodule, but multiple small bowel strictures may also be found, and sometimes reaching the jejunum [9].

Diaphragmatic involvement causes right basithoracic and scapular pain, sometimes simulating biliary colic.

Involvement of the umbilicus is rare.It is manifested by induration of the umbilicus at the time of menstruation and even umbilical discharge.

The intestinal involvement of endometriosis is generally linked to infertility[1].

In addition to these classic gastrointestinal disorders, functional bowel disorders are also observed, and irritable bowel syndrome is observed 2.5 to 3.5 times more frequently in women with endometriosis than in the general female population[10].

Despite the severity of this symptomatology, gastrointestinal endometriosis has long remained unrecognized, so that the diagnostic delay between the appearance of these symptoms and the diagnosis of endometriosis varies from 3 to 6 years.

The cyclical nature of the abdominal pain should lead to the diagnosis of endometriosis.

Preoperative evaluation
In the presence of endometriosis, certain tests are required.

The elevation of plasma concentration of CA-125 for the diagnosis of endometriosis at all stages: sensitivity at 30% and specificity at 90%. However, it is correlated with the severity of the disease [11].

The two most effective imaging tests for endometriosis are the endovaginalultrasound and the MRI, but these examinations are operator dependent [2],[6].

- Endovaginal ultrasound: this shows the depth of invasion of the vagina, bladder, and rectum by endometriosis.
- Endoscopic ultrasound: transrectal ultrasound gives the same informationas the endovaginal ultrasound on the invasion of adjacent pelvic organs by endometriosis.

- Colonoscopy: is carried out in case of rectal bleeding to search formalignantneoplasms. In the case of intestinal involvement, the submucosa is only affected in 30 to 40% of cases, and deeper in only 20% of cases. Involvement of

the mucosa, which is very rare (10%), can be easily and only be identified during menstruation. During the rest of the cycle, it may go unnoticed, as the observed abnormalities are covered by normal mucosa [2].
-CT scan: may sometimes show small bowel lesions in favor of endometriosis or can confirm bowel obstruction.
-MRI: can better characterize pelvic and abdominal organs affected by endometriosis.

Surgical Treatment
The prevalence of asymptomatic endometriosis discovered during laparoscopy varies from 6 to 50% depending on the studies and the indication for exploration [2].

In case of emergency, the treatment of ileo-caecal endometriosis is surgical,it consists of resecting the intestinal segment affected by the nodule either by laparotomy or by laparoscopy. Thus, by performing an ileo-caecal resection and ileo-colic anastomosis [5].

In the case of our patient, we performed an ileo-caecal resection removing both endometriosis nodules with a manual extra corporal side to side ileo-colic anastomosis. It was a radical intervention to prevent any risk of recurrence.

In certain situations where the endometriosis is in its early stages and is discovered during the operation, some authors use the "shaving" technique (resection from the outside inwards of the nodule penetrating the muscularis without resection of the submucosa).

Iconography :

Figure 1:- Plain abdominal X-ray, hydro-aeric levels.

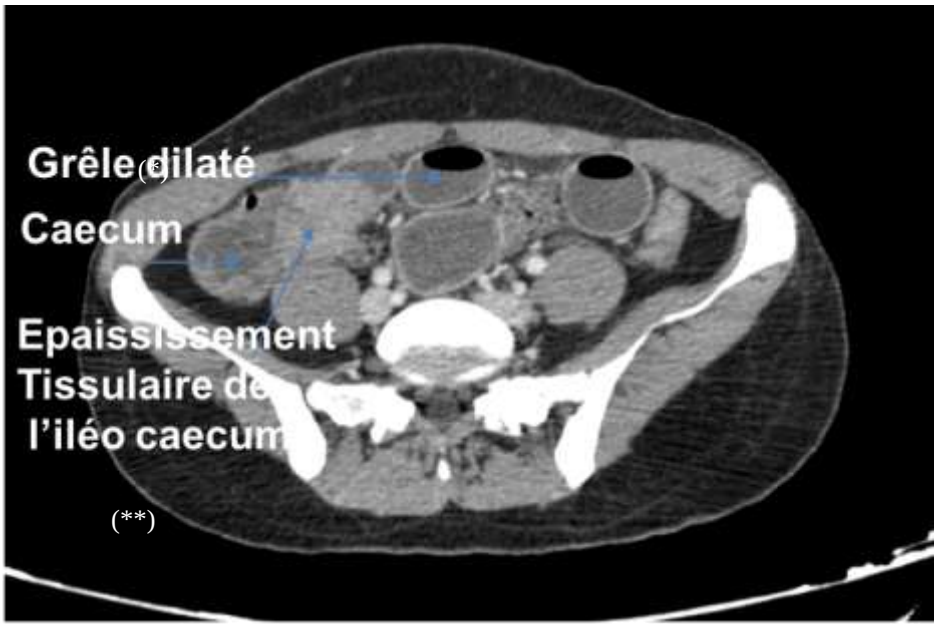

Figure 2:- Axial plane CT scan showing ileo-caecal stenosis with upstream dilatation.
(*) : small bowel dilatation
(**) :Ileocaecal thickening

Figure 3:- CT scan, coronal plane showing ileal stenosis with upstream dilatation.
(*) :Ileocaecal thickening

Figure 4:- CT scan, coronal plane showing a second tumor of the terminal ileum.

Figure 5:- Position of the four trocars.

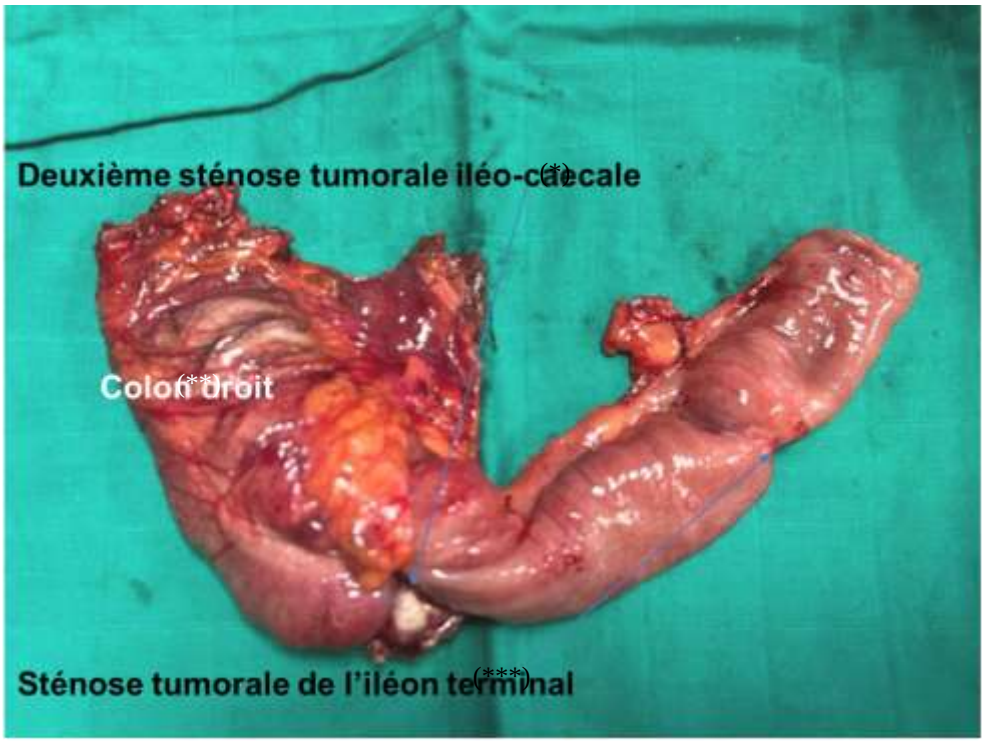

Figure 6:- Surgical specimen, ileo-caecal resection with lymph node dissection.
(*) : Ileo-caecal nodule
(**) : ascending colon
(***) : Terminal ileum nodule

Acronyms :
-CEA:Carcino embryonic antigen
-AFP:alpha foeto protein
-BMI: Body mass index
-CA 19-9 :Cancer antigen 19-9
- CA-125 : Cancer antigen 125
-dl :deciliter
-g : gram
-CRP :C Reactive Protein
-PPI :proton pump inhibitor
-mg : milligram
-mm : millimeter
- µg :microgram
- ml :milliliter
- L : liter
- U/L : unity per liter
- MRI : magnetic resonance imaging
-CT : computed tomography

Conclusion:-

Ileo-caecal endometriosis is rare and most often asymptomatic and difficult to diagnose because its main symptoms, in particular dysmenorrhea and infertility, are not specific and are often neglected or minimized.

It is then diagnosed during an abdominal surgery, by the discovery of small violaceous peritoneal nodules or in case of bowel obstruction episodes which are often incomplete and iterative, or gastrointestinal bleeding.

The treatment of ileo-caecal endometriosis is mainly surgical, aiming at resecting all the lesions.

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