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A Case Report of Extrapelvic Multilocular Endometriosis

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Abstract: ***Background: The prevalence of intestinal involvement of endometriosis is reported to occur in the literature 3-37 % of the diagnosed cases of extrapelvic endometriosis. The most common site is the recto-sigmoidien segment. The clinical complaint of intestinal endometriosis vary from abdominal mass and pain, to constipation, diarrhea, make it difficult to diagnose a neoplasic or inflammatory process with endometriosis, even with the help of imaging exams. The histopathological diagnosis could be challenging when we consider the metaplasia, hormonal and inflammatory changes, or the suspect of a carcinoma envolving the intestine. Case report: We will report the case of a 27 years woman that was diagnosed with an abdominal mass probably neoplasic that involve the rectum, small intestine, ovary and Fallopian tube, the hystopathological exam revealed an extrapelvic multilocular endometriosis. Conclusion: The diagnosis of endometriosis could only be confirmed by histopathologic examination, even there could be a lot of challenges since the alterations that associate this disease may mimic a wide range of disease.***

Keywords: endometriosis, intestinal endometriosis, endometroid carcinoma

1. Introduction

Endometriosis is the implantation of endometrial tissue outside the uterus, usually it is composed of glands and stroma of endometrial tissue, but it can be only glands or only stroma.

The prevalence of endometriosis reported in the literature is (3)

1-2 %	among woman infertile age
15-20%	Among woman with infertility problems
40-60 %	Woman with dysmenorhea

It was usually classified as a benign diseases although its behavior typically is neoplasic. Generally it is found in the surface of the ovary, fallopian tube, bowel, urinary tract, but there are presented also rare cases of pulmonary, cerebral, musculary sites. [6 7 8]. There have been report also some rare cases of endometriosis in men with estromimetic hormonal treatment [9]. Although there are not resumptive studies, the most common extrapelvic sites reported are abdominal scars (11.43%) [5] and gastrointestinal ones with a incidence from 3-37% of the diagnosed cases [4], with the rectosigmoidien being the most common site of involvement.

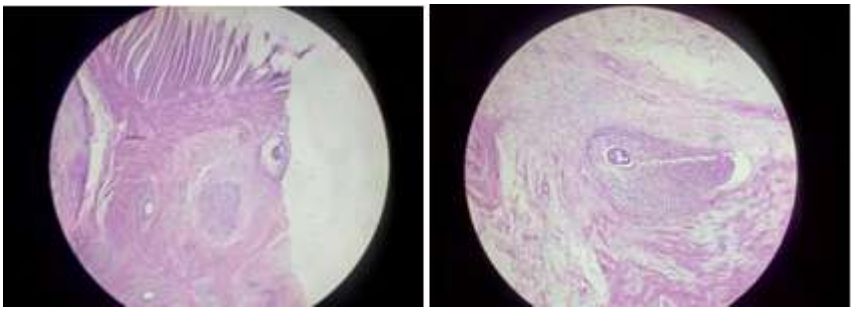
The most common complaints included abdominal pain, an abdominal mass, intestinal obstruction, rectal bleeding, infertility, diarrhea and increasing urinary frequency [10]. The severity of the disease depends on the depth of the involvement of the bowel wall.

2. Case Report

The patient B.M 27 years old, presented at the emergency room with severe abdominal pain and intestinal obstruction. She refers that have had this symptom for several times, but lately they were getting worse. She did not refer about rectal bleeding or periodic appearance of the symptoms. On the physical examination she looked pale, the abdomen was tender and painful and an abdominal mass was found. An ultrasonographic examination was performed that revealed an abdominal mass involving the ovaries, the rectum and some segments of small intestine. The imagery examination did not offer any further information, so an exploratory laparoscopy was performed followed by a radical chirurgical procedure including ovaries, rectum and a 10 cm segment of small intestine were excited for an abdominal neoplasia.

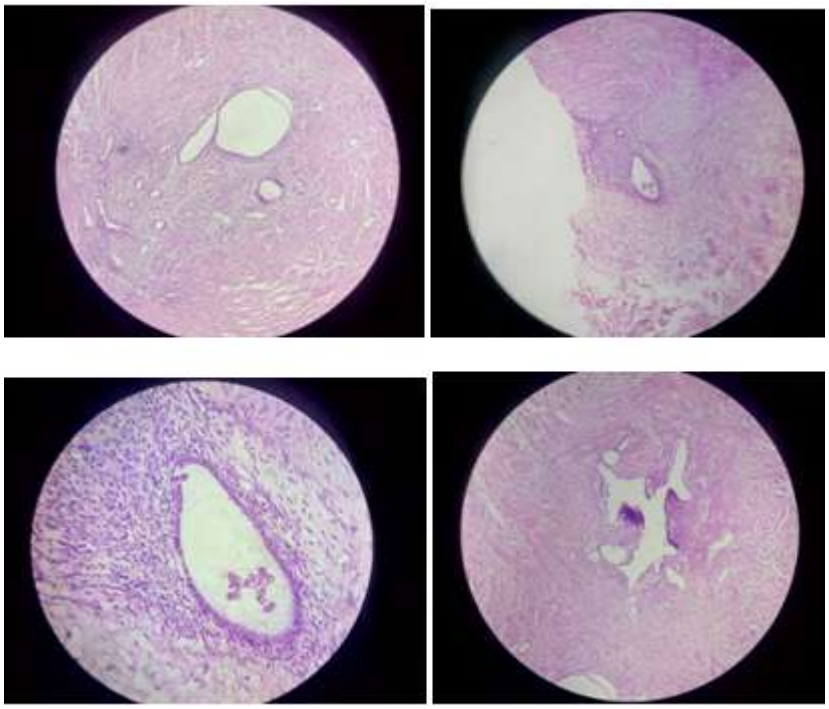
The histopathological macroscopically examination shows that the muscularis propria of the rectum was infiltrated by a formation with no evident margins that did not affect the lumen with segments of hemorrhage and the wall of rectum was firm and thick. The mucosa looked intact. The same lesion was found on the small intestinal segment associated with fibrosis. The surface of the ovaries was irregular, with foci of fibrosis and hemorrhage.

Microscopically on the wall of rectum in the middle of muscle fibers were found glands and stroma similar to those of the endometrial tissue, some of the section present only stroma, we found also foci of hemorrhage and fibrosis, inflammatory infiltrate. Some of the glands represent epithelial hyperplasia without atypia, tubal metaplasia were found, other were dilated covered by flat epithelial layer, presenting some focuses of hiperchromasia. The same lesions were found on the sections from the ovary. *[picture 1-8]*



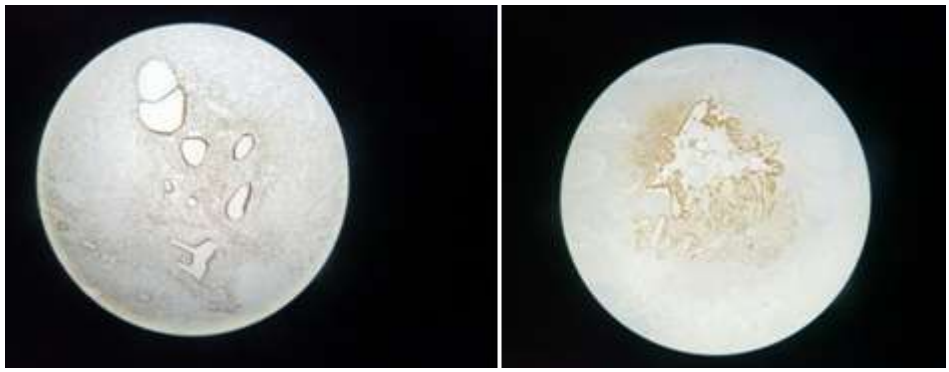
Picture 1-2 . HE stain: in the middle of intestinal muscle fibers there were focuses of stromal or glands and stroma similar to endometrial tissue.

Picture 3-8 HE stain: hemorrhage and fibrosis, inflammatory infiltrate around the glands Some of them represent epithelial hyperplasia without atypia, tubal metaplasia and hyperchromasia.

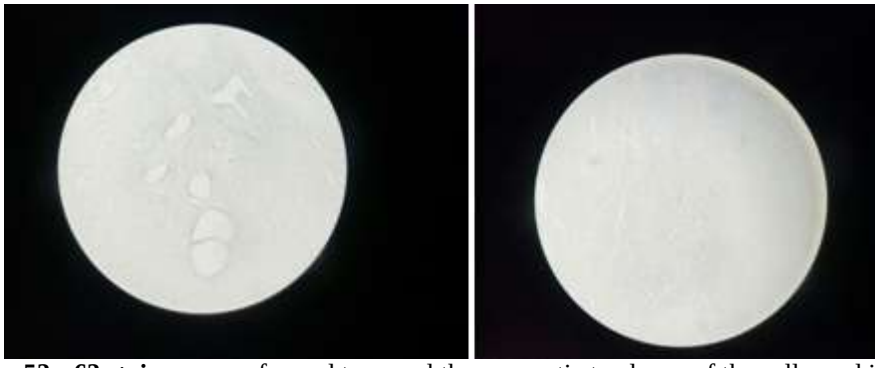


The imunohystochemistry was performed and the results were ER (+) *[picture 11-12*] ;CD 10 (+) *[picture 9-10]* p53 (-) p63 (-) *[picture 13-14*] confirming the origin of the infiltrated tissue from the endometrium, and the negative oncocytic behavior of the cells, leading to the diagnosis of extrapelvic multilocular endometriosis.

Picture 9-10 CD 10 stain: was performed to reveal the endometrial stroma and as we see in the picture it was positive



Picture 11-12 ER stain: was performed to confirm the endometrial origin of the tissue and it was positive on the epithelial and stromal tissue



Picture 13-14: p53-p63 stain: was performed to reveal the oncocytic tendency of the cells, and it was negative.

3. Discussion

Endometriosis is defined as endometrial glands and/or stroma outside the endometrial cavity [11]. Recently studies shows that extapelvic endometriosis occupy from 4-12 % of histology proven endometriosis [24,28], one third of which are intestinal ones [4]. The frequency of this disease in the several parts of the bowel was 39% in the sigmoid colon , 20% in the recto-sigmoid colon , 19% in the appendix , 10% in the rectum, 7% in the terminal ileum, 4.5% in the caecum and 0.6% in the transverse colon : sigmoid and rectal endometriosis accounted for 69% of the cases [1,2,3,4,26]. In all this range of distribution, as a special task, the diagnosis of multifocal endometriosis, even though is not common, it has been documented [26] and must be taken into consideration.

Although the ethiology and fispathology of endometriosis, is not totally clear, Sampsons for the first time described various theories that some time on their one, and some time in combination with each-other try to explain the fispathology of endometriosis [12]

- The metaplasic theory that support the metaplasia of residual Mullerian tissue
- The implantation theory that support the displacement of endometrial tissue in different ways, lymphogenic, vascular, retrograde menstruation, physical implantation during surgical procedures ect.
- The combination of both theories

Recent studies support all three theories, modified them with the genetic factor and alterations of the immune system that allow the endometrial cell to survive on the body [13].

Clinically it is represent with a wide range of complaint, that in intestinal endometriosis endometriosis vary from abdominal mass and pain, to constipation, diarrhea, symptoms of sub acute or acute intestinal obstruction [26,27]. This symptoms only on 41% of cases is

characterized by cyclic appearance, make it difficult to diagnose a neoplasic or inflammatory process with endometriosis. Some special studies have present the involvement of mucous, making the diagnosis more difficult [15,21]. When the mucous is involve the clinic became more complicate gastrointestinal bleeding, rectal urgency ect [15,20,21,23,25,27,20]

In this clinical context the first differential diagnosis of the intestinal endometriosis is a carcinoma, primarily an intestinal adenocarcinoma, but also metastasis for other carcinomas.

Imaging exams can help and direct the diagnosis, however, none of these exams is capable of confirming the diagnosis *per se.* The endoscopic biopsy can't arrive till the muscularis propria, so it can't give us any information. Endoscopy can be useful on the cases present with popypoid intraluminal mass or annular lesions [21]. It can be identified on double barium enema as a extrinsic mass, but it can't give us any further orientation [29]. Us and CT can only identify the thickens of the intestinal wall, but recently devised methods such as endoscopic US and CT (virtual) colonoscopy can also be used in the diagnosis of endometriosis seen in the recto-sigmoid region [17,19,26,29] . The gold line standard for the diagnosis is laparoscopy with histopathology [14].

When it comes to histopathological diagnosis, on hematoxilin-eosin stains typically finding the edometroid gland and stroma make the diagnosis clear, but with all the metaplasia, hormonal, and inflammatory changes that affect the endometroid tissue, it is difficult even histologicaly to differ an endometroid carcinoma involving the intestinal tissue, from pure endometriosis. Typical cases showed stroma cuffing around the endometrial glands [18, 22,23,25,26]. The case we report histologicaly presents the endometrial gland and stroma, but we found focuses of only glands and only stromas, with metaplasia and inflammatory changes.

The diagnosis of Intestinal Adenocarcinoma it is usually taken into consideration when the mucous is involved in endometriosis [16,20,22,23], and in this cases it clinically associated with rectal bleeding.this cases become more complicated when atypical hyperplasia involve the glandular component, or the stromal component is undergoing mixoid alteration. All this changes can lead to misdiagnosis of intestinal adenocarcinoma. On this cases the use of the immunohystochemistry is necessary for the diagnosis CK 7 +/ CK 20 – dhe ER (+), sometimes accompanied with CD 10 + direct to award the endometrial tissue. [18,19]

The diagnosis becomes more difficult when we consider the malignant transformation of intestinal endometriosis [20,22], so squamous, eosinophylic and mucinous metaplasia features should make us search for a typical endometrial gland to direct the diagnosis, and for definitive diagnosis the immunohistochemistry must be undertaken [18,19,20,22]. In this prespective the use of p53/p63 markers can guide us to oncotic transformation of the cell [18,19,22,23,30,31,32]. In our case we tested the oncotic transformation by using the p53/p63 markers.

Primary endometrial carcinoma of endometriosis must be taken into consideration when we see complex hyperplasia with atypia [22,23].

In some other cases the parietal intestinal endometriosis, in particular in multilocular endometriosis, the differential diagnosis of metastasis from endometrial adenocarcinoma (mostly Figo I/II) is the first to be excluded. The immunostain markers are useful [30, 31,32]

Endometrial stroma may have a variety of changes including decidua, pseudodecidua, smooth-muscle metaplasia, fibroblastic metaplasia, and sarcoma.

4. Conclusion

In summary the presence of endometriosis can be predicted based on history and physical examination by the clinians. Although the gold line standard for a proper diagnosis is histopathologic examination, the pathologist could run into a lot of challenges since the alterations that associate this disease may mimic a wide range of disease.

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