Case Report

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Ganglionic endometriosis with primary focus on the rectum–a rare case report

Abstract

Endometriosis is a pathology that is increasingly been diagnosed in women of reproductive age. It is located essentially in the pelvic region but may have an extrapelvic location. We describe a rare and exceptional case of an incidental intraoperative finding of a rectal stenotic mass, whose further study revealed an endometriosis focus with ganglionic extension to mesorectum.

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Introduction

Endometriosis is a clinical entity defined by the presence of functioning endometrial tissue at sites outside the uterus.[1,2] It mainly affects women at fertile age in about 6-10% and is usually located on the peritoneal surface of the reproductive organs. Extrapelvic involvement like rectosigmoid junction, sigmoid colon and rectum is very uncommon. Symptoms such as pelvic pain, infertility or both are described in 40-80% of patients.[2,3] Colorectal endometriosis can be asymptomatic or produce non-specific symptoms such, pelvic pain exacerbated with menstruation, rectal bleeding, dyspareunia, pain on defecation, constipation and bowel obstruction.[4] Circumferential involvement of the rectum is rare and should be differentially diagnosed from inflammatory or malignant diseases.[4,5]

We report a very rare case of an obstructive rectal mass of endometriosis with ganglionic involvement.

Case report

We report a case of a 41-year-old woman with an history of chronic anemia and dysmenorrhea which was submitted to total abdominal hysterectomy and left-sided annexectomy right ovary preservation. During the procedure, a stenosing lesion of the rectosigmoid transition was observed. No approach to that lesion was taken because further examinations were required. After clinical history, it was found an history of constipation over the last weeks and an abundant rectal bleeding without hemodynamic compromise, colic abdominal pain and some abdominal distension mainly during menstruation. As pathological antecedents, we mention Otto's disease (it contraindicates hormone therapy), congenital hip dysplasia and two transient ischemic vascular accidents. She was taking clopidogrel and aspirin and has no family antecedents. Some studies were performed. Complete blood tests and tumor markers were normal. A total colonoscopy was performed which identified a stenotic narrowing (15.8mm non-franchise at the 12.8mm colonoscope) at 15cm from the anal margin, where the mucosa presented a discreetly infiltrative / spongy appearance but with no inflammatory features. The suspicion was endometriosis. Biopsies of the lesion were performed but the histology was inconclusive only identifying inflammatory infiltrate, edema, and epithelial aspects of the regenerative, peudopolipoid type. It was not found any criteria for morphological or pathological diagnosis. Six months later, she repeated the colonoscopy in which it was identified an insurmountable stenosing lesion whose mucosa appeared normal. Newer biopsies were also inconclusive. She made a complete body tomography reporting a thickening in the rectosigmoid transition without adenopathies or other relevant findings. Two months later she undergone to a segmental resection of the rectum with total mesorectum excision without intercurrences. She was discharged on the 6th postoperative day without pain and normal bowel function was maintained without blood losses. The histological diagnosis confirmed the endometriosis of the rectum with involvement of 23 lymph nodes. At present moment, the patient is well without any complaint and has gynecological follow-ups.

Discussion

Endometriosis was first described as the presence of functioning endometrial glands and stroma outsider the uterine cavity. As stated previously, women of reproductive age are affected in about 6-10% complaining of pelvic pain and infertility in a large number of female patients. Bowel obstruction occurs in 10% of all cases of endometriosis.[6] colorectal obstructive endometriosis with colon or rectum involvement is rare. Over the years many theories have attempted to explain endometriosis pathogenesis such as Sampson´s theory of retrograde spread, vascular dissemination, autoimmune disease, colonic metaplasia among other.[7] Abros et cols suggest that endometrial tissue which grows around the rectum, and obstructs more than 80% of lumen are more able to infiltrate the nearest ganglions.[8] Some authors point out two hypotheses to explain lymphatic affection such as metaplasia process of secondary Mullerian system and lymphatic drainage of endometrial tissue, defined at five different routes.[9,10] Intestinal endometriosis symptoms vary according to the site of involvement. Abdominal pain, nausea, vomiting, fecal tenesmus, painful defecation, alternating constipation and diarrhea distention and rectal bleeding. In our case, the patient complaint about

bowel habits changing, rectal bleeding and abdominal pain which increases during menstrual period. Gastrointestinal endometriosis (3-37% of an ectopic location) may affect the ileum, appendix, sigmoid colon and rectum being more frequent in the rectosigmoid (50-90%) colon. Intestinal endometriosis often presents as a sub-mucosal tumor or luminal stenosis, because it mainly involves the muscularis propria and subserosa or even mesentery. Preoperative diagnosis of colorectal obstructive endometriosis is often difficult because of lack of definitive diagnostic, clinical and radiologic findings. The case described emphasizes the incidental finding during a hysterectomy of a lesion which the main concern was a malignant rectal lesion. After several radiological exams including biopsies, the diagnosis remains doubtful. As we far know, patients with chronic symptoms is easier to make a connection and the study should include computed tomography (CT) scans, magnetic resonance imaging (MRI), and positron emission tomography (PET) scan in order to avoid false positive diagnosis of malignant tumors. MRI appear to be more sensitive technique for colorectal endometriosis with a positive predictive value about 89%. Endoscopy is another tool used in the diagnosis specially for making biopsies. However, even with mucosal involvement, a clear diagnosis cannot usually be provided, because most of the times only shows some unspecific inflammatory infiltration, as reported in our patient´s case. It can be justified because the histological bowel wall changes involving endometriosis are located between muscularis fibers, subserosa, and serosa, but mucosa is almost always intact. From the recent literature, laparoscopy diagnosis is now the gold standard approach to diagnosis and the histopathologic and immunochemistry study confirms it. Colorectal endometriosis treatment is usually conservative (hormone therapy) in order to relief the symptoms. Therefore obstruction, bleeding or no medical response requires surgical procedures. Free surgical margins are an important step after the surgical removal because it is difficult to differentiate or exclude malignancy. Of the available literature, very little is known about the most appropriate diagnosis for the affected nodes, nor is their approach, consequences, clinical evolution or more adequate treatment. Further studies are needed to better understand this issue.

Conclusion

Colorectal obstructive endometriosis is rare and ganglion involvement is so far much rare. We should be aware of the symptoms in females in reproductive age specially the ones which became worst during the menstrual period. Imaging exams could help but most of the time is inconclusive and cam mimic other pathologies. Laparoscopy diagnosis is a gold tool and histopathology confirms the diagnosis after tissue removal. Malignancy should always be excluded. The lymph node spread, its treatment, evolution and prognosis is still a limited field that requires more studies for better understanding and approach.

Acknowledgment

None.

Conflict of interest

The authors declare no conflict of interest.

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