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ENDOMETRIOSIS OF APPENDIX – AN INCIDENTAL FINDING

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ABSTRACT
Pain in the right in iliac fossa can be a related to organ localized within this quadrant. The most appropriate clinical diagnosis for tenderness in McBurney's point is acute appendicitis. An uncommon cause for pain in the right iliac fossa is endometriosis of appendix. We document a case of 35 year old lady who presented with menorrhagia and bilateral ovarian cysts with adherent appendix to (Rt) ovary. Histopathological examination of surgically excised specimen confirmed it to be bilateral ovarian endometriosis with endometriosis of appendix. This case highlights difficulty in making preoperative diagnosis in this uncommon entity and importance of specimen sampling and histomorphology of appendix for rendering the right diagnosis. In addition, differential diagnosis of endometriosis of appendix should be considered as one of the rare possibility in women particularly when pain is related to menstrual cycle.

KEYWORDS: Appendix, Endometriosis.

INTRODUCTION
Pain in the right iliac fossa may be attributed to various conditions with acute appendicitis being the most common etiology. Other causes include amoebic typhilitis, inflammatory bowel disease, ilieo-caecal tuberculosis, inflammed solitary diverticulum of the caecum, ectopic pregnancy and pelvic inflammatory disease in women. In addition to all these, an uncommon etiology of right iliac fossa pain may be an appendicular or caecal endometriosis. A clinical diagnosis of appendicular endometriosis is usually not made pre-operatively which in most instances revealed only after histopathological examination of appendix removed for suspected appendicitis. In this case report, we document a case of appendicular endometriosis, which was diagnosed clinically as bilateral ovarian cysts.

Clinical presentation of endometriosis varies, and some affected women may be asymptomatic. Symptomatic women have mild to severe pain that may or may not be cyclic depending on the site involved and severity of the disease. Endometriosis most often causes chronic pelvic pain; however, women may suffer acute abdominal or pelvic pain severe enough to cause them to seek emergency medical care.[1]

Appendiceal endometriosis was first described in 1860.[2] Endometriosis of the appendix is uncommon, and presentation as acute abdominal pain is rare. Endometriosis of the appendix may be asymptomatic or present as acute appendicitis, lower gastrointestinal bleeding, intestinal perforation, or intestinal obstruction from intussusception.[1-2]

The aim of this case report was to describe our experience of managing patients with appendiceal endometriosis and to review the clinical characteristics of this medical condition.[2]

CASE REPORT
A 35-year-old lady was admitted with history of recurrent lower abdominal pain and menorrhagia not responding to hormones, for endometrial evaluation. She had single male child, 10 years of age with normal delivery. There was no significant past medical history. Her menstrual cycles were normal with normal flow. On general physical examination, patient was afebrile and vital parameters were normal. Per abdomen examination revealed tenderness in pelvic region.Ultrasonography of abdomen and pelvis showed features of bilateral ovarian cysts favouring endometriosis. Endometrial thickness was 8 mm and rest within normal limits. CA 125 markers were within normal limits. Bilateral ovarian cystectomy was planned and after fitness she was posted for the same. During surgery it was found that both ovaries had large unilocular cysts with (Rt) ovary adherent to appendix. Bilateral ovarian cystectomy was done along with (Rt) sided partial ovarian excision and appendicectomy. Excised ovarian cysts and appendix were sent for histopathological examination.

Postoperative period was uneventful and she was discharged on third postoperative day. She is on regular follow up.

Morphology

Received two specimen labeled as (Lt) ovarian cyst and (Rt) ovarian cyst with appendix. The specimen of appendix was 10 cm in length, thickened, tip was enlarged. Cut surface revealed totally obliterated lumen and thickened serosa. Both ovarian cysts were 3x2 cm already cut opened, unilocular with smooth wall.

Microscopic Features

Sections from bilateral ovarian cysts showed ovarian stroma along with round tubular endometrial glands in fibrous stroma. There was no evidence of hyperplasia or malignancy. Section from Appendix stained with H & E showed totally obliterated appendicular lumen, mucosa and beneath fibrocollagenous stroma along with scattered lymphocytic collection and hypertrophied muscular layer showing few round tubular endometrial glands in low power. (Fig. 1) and seen prominently in(fig. 2).

Hence the diagnosis of bilateral ovarian endometriosis with endometriosis of appendix was made.

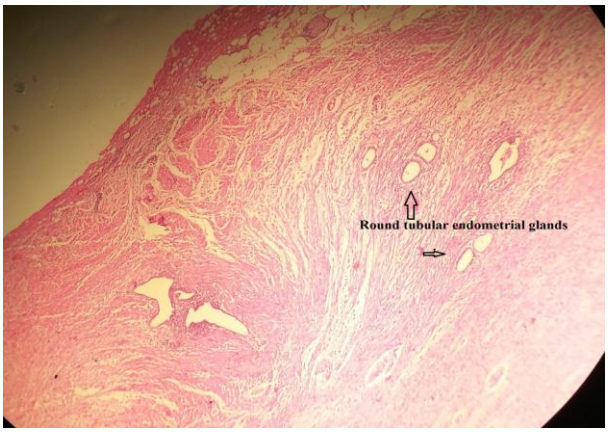

Fig. 1: Showing Round Tubular Endometrial Glands With Stroma In Hyperplastic Muscularis (H & E 10x X 10x).

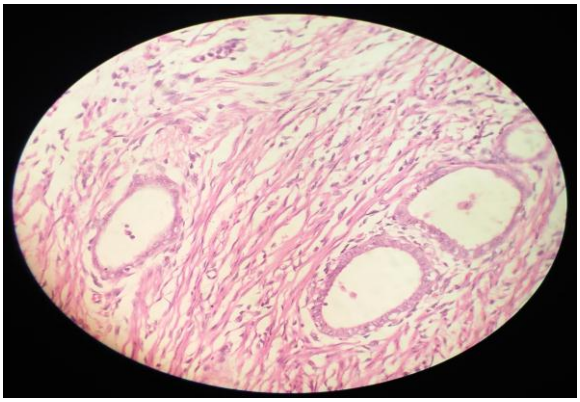

Fig. 2: Shows High Power View Of Fig 1 (H & E 10x X 40x).

DISCUSSION

Endometriosis is an estrogen-dependent lesion, occurring commonly in young women, featuring endometrial glands and stromal tissue in extra-uterine location. Its incidence ranges from 4 to 17% among women of reproductive age group and usually affects females of 25 to 45 years of age.[1] Common sites of this entity with presence of ectopic endometrial tissue includes ovaries, peritoneum, uterosacral ligaments and rectovaginal cul-de-sac. While involvement of appendix is extremely rare and can mimic acute appendicitis.[1-2] The main symptom of endometriosis is chronic abdominal pain, and the diagnosis is often made later, mostly after the result of the histopathological examination. Endometriosis of the cecal appendix is rare and almost never diagnosed before the surgery[2], with the definitive diagnosis obtained only after microscopic examination of tissue. However, it should always be taken into account for the diagnosis of chronic pelvic pain, especially in young women complaining of recurrent pain, history of infertility and pelvic endometriosis.[2]

Among the most accepted theories of endometriosis, coelomic metaplasia is mentioned, as well as retrograde menstruation, proposed by Sampson, and the genetic theory, besides blood and lymphatic spread. Review of literature reveals that the incidence of involvement of the gastrointestinal tract varies from 15 to 37% of the cases and it most commonly affects the rectum and the recto-sigmoid junction accounting for 72 % of the cases.The small intestine involvement is around 7% more likely restricted to the terminal ileum. The less affected sites are the cecum and appendix. Several studies focused on appendicular endometriosis case series show incidence of < 1%.[1-2]

Appendiceal endometriosis is classified into primary and secondary types. Lesion with histopathological evidence of endometriosis within the appendix with no clinico-pathological evidence of extra-appendicular endometriosis is described as primary form while secondary form is associated with features of external endometriosis. However, primary appendiceal endometriosis cases have also been reported without involvement of any other gynecological organ.

Based on clinical symptoms, appendiceal endometriosis may be sub classified into four groups: (1) Patient presenting clinically as acute appendicitis; (2) Patient with appendix invagination; (3) Patients presenting with non-specific symptoms like colicky abdominal pain, nausea/vomiting and malena; (4) Cases who are asymptomatic.[1] Finding of an inverted or bulbous appendiceal orifice on colonoscopy suggests a more likely diagnosis of appendiceal endometriosis. Appendiceal wall affected by endometriosis show secondary changes such as chronic inflammation, fibrosis, hypertrophy/hyperplasia of muscularis propria. Presently there is no investigation that can give definite diagnosis of endometriosis pre-operatively. Radio-imaging may also be of not much help for its diagnosis.

Although, laproscopy, may be considered as a standard method for diagnosis as features visualized under magnification may give direct clue. However, variable lesions may be noted limiting its importance. Common locations of endometriosis in appendix are tip and body. In a study on appendiceal endometriosis by Mittal et al, about 56% showed involvement of body and 44% tip of the appendix while base of the appendix was not involved in any of their cases.[1] A definitive diagnosis of endometriosis can only be given on histopathology. The histologic hallmark of endometriosis includes presence of endometrial glands, stroma, fibrosis, and hemosiderin-laden macrophages. Layers involved by endometriosis of appendix wall include serosa and seromuscular. In the present case, the lesion was localized to tip of appendix and involved serosa and muscularis propria. It becomes a challenging task for a pathologist when endometrial glands are absent or sparse with evidence of may be stroma, which can be termed as stromal endometriosis., which can be filled with erythrocytes as noted in our case. Management of endometriosis of appendix depends on presentation. Symptomatic cases presenting with pain is the most common indication for surgical intervention. Pathologist should be aware of the fact that if appendectomy specimen received for a clinical diagnosis of acute appendicitis, does not show microscopic features of acute appendicitis, adequate sampling/ all embedding of appendix should be done to rule out acute appendicitis or any other etiology and even a rare possibility of endometriosis.

In a known case of endometriosis, pain in abdomen and pelvic region a diagnosis of endometriosis affecting the specific tissue involved should be considered. This diagnosis should be more strongly suspected clinically, if the pain corresponds to the menstrual cycle. Appendiceal endometriosis appears to be an incidental finding, and one that is clinically less thought of.

In conclusion, appendiceal endometriosis is rare, and its preoperative diagnosis is difficult. However, it should be included in the differential diagnosis of acute abdominal pain or menorrhagia, especially in women of child bearing age.

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