

# Rare extrapelvic endometriosis on iliac vein wall – diagnosis and treatment

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## Summary

Although endometriosis is a relatively common illness in women during the reproductive period, extrapelvic localization of endometriosis is a relatively rare finding which can pose a differential diagnostic problem. Diagnosis and surgical treatment of a patient with endometriotic tumor located on the outer wall of the left common iliac vein are discussed. Patient underwent surgery because of circulatory disorders and left leg edema. Histopathological findings verified endometriosis on the outer wall of the common left iliac vein without other localizations.

*Key words:* Endometriosis; Iliac vein compression.

## Introduction

Endometriosis is defined as the occurrence outside the uterine cavity of areas of glandular structures surrounded by specific stroma, histologically and functionally resembling uterine endometrium. Although they may be found in any organ in the body, the lesions predominantly occur in the pelvic region, involving the ovaries, broad ligaments, rectovaginal septum, uterosacral ligaments, uterovesical pouch, the surface of uterus, and oviducts.

Localization of endometriosis can be divided into two broad anatomical categories – pelvic and extrapelvic. Pelvic lesions may be found within the ovaries, most frequently in fallopian tubes or pelvic peritoneum. Extrapelvic lesions may be localized to the genitalia, perineum, urinary tract system, gastrointestinal tract, intrathoracic structures, musculoskeletal tissue, abdominal wall or nervous system [1].

Endometriosis is an estrogen-dependent inflammatory process affecting 10% to 15% of fertile women, and as many as 50% of infertile women [1, 2]. As the disease is often asymptomatic and remains undiagnosed, its true incidence is not known. The diagnosis of extrapelvic endometriosis is difficult, usually dictated by clinical suspicion, followed by imaging studies (ultrasound, CT and magnetic resonance imaging), laparoscopy, tissue biopsy and immunohistochemistry [3, 4].

Due to high frequency of occurrence of endometriosis in women in reproductive period, especially in cases followed by cyclical repetition of discomforts, extrapelvic endometriosis should be taken into consideration regardless of numerous differential and diagnostic dilemmas. The aim is to achieve better preoperative preparation and to conduct operative procedure with minimum intraoperative and postoperative complications leading to better treatment prognosis.

## Case Report

Case is presented of a patient age 31 who was surgically treated for clinically diagnosed compressive tumor of left common iliac vein. After such treatment, extrapelvic endometriosis, i.e., endometriotic tumor on the outer wall of the left common iliac vein was histopathologically verified.

Discomforts which caused patient to report for checkup started one year before that. Swelling of left foot started first. Over time, the edema kept spreading upwards so after 6 months the entire left leg was swollen. That is when clinical suspicion arose differentially diagnostically either about left parailiac lymphadenopathy or deep vein thrombosis in iliacophemoral region. Color Doppler ultrasonographic examination was performed and clinically suspicion set about external compression of left common iliac vein. An MNR scan of the pelvis was performed because of suspected lymphadenopathy and it was ruled out. MNR scan showed that uterus and both ovaries were normal in size and shape which, together with previously performed detailed gynecological exam, confirmed absence of gynecological condition. Patient underwent surgery. Intraoperatively, a tumor lesion was found on the left common iliac vein which was the cause of compression; it was removed completely and sent to histopathological exam. Iliac vein was reconstructed by ringed tubular graft and an artificial arteriovenous fistula was made. Postoperative course was complicated on the second postoperative day by conspicuous edema of the left leg – clinical exam raised suspicion about thrombosis of the arteriovenous fistula, and the patient was operated again. Postoperative course was complicated again at first by fever and 5 days later by repeated occurrence of edema of the left leg. Checkup phlebography then showed vein graft thrombosis while blood flow in AV fistula was normal. Cavography showed normal flow through inferior vena cava, with no external compression. Anticoagulant, antibiotic and physical therapies were prescribed. Patient reacted well to administered therapy. After all discomforts and clinical symptoms of infection disappeared, and normal vein flow through the reconstructed iliac vein was confirmed, AV fistula ligation was performed and patient was released from hospital in good general state. Histopathological exam of the tumor surgically removed from the wall of the left common iliac vein verified endometriosis. Regardless of completely normal gynecological findings, i.e. no signs of any gynecological condition, regular checkups with vascular surgeon

Revised manuscript accepted for publication March 12, 2013

and with gynecologist were continued. All findings were normal. Patient was discomfort-free. The only problem that appeared later was sterility issue. In spite of completely normal gynecological finding, ovulatory cycles, normal hormonal status and husband's normal spermogram finding, patient was not able to conceive naturally through spontaneous cycle nor through stimulated cycle, or by any in vitro fertilization (IVF) method.

## Discussion

According to references, extrapelvic endometriosis is rare. The most common localizations of extrapelvic endometriosis are scars remaining after surgical procedures (episiotomy, Caesarian section) [2, 5], followed by gastrointestinal tract. The gastrointestinal tract is the most common site of extrapelvic endometriosis, affecting 5% - 15% of women with pelvic endometriosis. Among women with intestinal endometriosis rectum and sigmoid colon are the most commonly involved areas [6]. Inguinal endometriosis is rare and accounts for 0.3-0.6% of patients affected by endometriosis [7]. An isolated case of endometriosis of obturator nerve was reported in 1990 [8]. A unique case of retroperitoneal endometriosis causing deep vein thrombosis from extrinsic compression of the right iliac vein is described [1] as well as endometriosis causing cyclic compression of the right external iliac vein with cyclic edema of the right leg and thigh [9].

Also, there are two case reports of endometriosis occurring around large veins. The first case reported endometriosis involving the left femoral vein [10]. The patient presented with a painful groin mass that showed characteristics of endometriosis in the adventitial layer of the vein. The second case was due to endometriosis encircling the right external iliac vein causing edema of the right leg and thigh. Successful surgical resection of the endometriosis was performed in both cases [9].

Management of endometriosis is individualized and driven by lesion localization and symptoms presentation [2, 11]. Considering that endometriosis relapse is a frequent finding, especially in cases of pelvic localization, and that remission depends on adequate and complete surgical removal of endometriotic tumor; adequate surgical intervention is necessary in order to prevent relapses especially in cases of extrapelvic localization. However, rare extrapelvic localization of endometriosis and varying symptomatology is often the cause of erroneous preoperative diagnosis.

Sterility poses as a particular problem and occurs in about 50% of patients suffering from extrapelvic endometriosis even after it is successfully surgically removed as in this case.

The role of endometriosis in development of infertility (except in cases of mechanical obstruction to conception) is still unknown. Considering the fact that some patients with this condition conceive without any difficulties, while other are infertile, it is clear that there is no unique explanation for all infertility cases.

Question whether completely removed extrapelvic endometriosis should be postoperatively treated by medications, which medications should be used and for how long, i.e., whether such a treatment of adequately surgically removed extrapelvic endometriosis can later affect reproductive ability i.e. fertility, still remains open.

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