# CYSTIC LYMPHANGIOMA ARISING FROM THE SMALL INTESTINE MESENTERY INCIDENTALLY FOUND DURING SURGERY FOR A LARGE OVARIAN TUMOR – A CASE REPORT

DOI: 10.36740/WLek202209122

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#### ABSTRACT

The aim of this study was presenting significance of diagnostic process in doctor's daily routine. A 45-year-old patient for a planned laparotomy due to left ovarian cyst detected with a routine transvaginal ultrasound. She did not report any symptoms. Computer tomography of the abdomen and pelvis showed a cystic lesion with segmental wall thickening, measuring 133 x 83 x 135 mm, adjacent to the left ovary and the uterus on the left side. At the laparotomy exploration, a giant cyst, ca. 20 cm in diameter was found above the uterus, on the left side, in the area between the lower and middle abdomen. The histological exam results showed lymphangioma of the small and large intestine mesentery. In this case, lymphangioma was found in a rare location, in the mesentery of the ileocecal valve. It did not present any signs and symptoms. CT used in the present case failed to identify the exact point of origin of the lesion.

KEY WORDS: cyst, lymphangioma, mesenteric cyst, small intestine mesentery

Wiad Lek. 2022;75(9 p1):2170-2173

# INTRODUCTION

Lymphangiomas are benign, non-epithelial cysts composed of lymph vessels [1]. They are most common in children and the majority are congenital malformations diagnosed before the age of 2 years [2]. Lymphangiomas are usually found in the face, back and front of the neck or in the axillary region [1]. They have been estimated to account for 50% of all mesenteric cysts [3] which occur with an incidence of 1:100 000 to 1:250 000 surgical admissions [4]. These estimates are based on single case reports of mostly incidentally discovered lymphangioma. Lymphangiomas are usually asymptomatic and any issues that may arise are associated with their atypical location, a large size and extent of the disease. We present a case of mesenteric lymphangioma of the small and large intestine which despite its location did not produce any symptoms or affect the patient's quality of life.

#### THE AIM

The aim of this study was to present the significance of the diagnostic process in doctors daily routine. In this case we did not use all of the available methods. Before the operation it seemed that the problem was evident and common. We would like to present that doing additional examinations is never an unnecessary diagnostics.

### **CASE REPORT**

A 45-year-old patient, gravida 1, para 1 (vaginal delivery) was admitted to the Department of Gynecology and Obstetrics, Solec Hospital in Warsaw for a planned laparotomy to remove a left ovarian cyst detected on routine transvaginal ultrasound.

Her general health was good and she was not on any medications. She had regular menses and denied any abdominal pain, abdominal distension, constipation or difficulty passing stools or urinating.

15 years prior to the current admission she had a laparotomy for a suspected right ovarian cyst and an umbilical mass. Partial bilateral ovariotomy was performed with the removal of the ovarian cyst and the umbilical mass, which was diagnosed by histopathology as umbilical endometriosis. The histopathological assessment of the ovarian specimens was not available.

On admission, an abdominal ultrasound identified the probable point of origin of the cyst in the left ovary. A bimanual recto-vaginal examination revealed an anteverted, mobile uterus of normal size. An ill-defined, non-tender, slightly mobile mass, approximately 15 cm in diameter, was found in the left adnexa. The right adnexa was unremarkable and non-tender. A pelvic ultrasound revealed a lesion in the left adnexa, measuring 60 x 50 mm, showing



Fig. 1. CT scan of the uterus



**Fig. 2.** CT demonstrating a cystic lesion displacing the mesentery in an upward direction.



**Fig. 3.** Intraoperative photograph of the mesenteric cyst of the small and large intestine.



Fig. 4. Lymphangioma (hematoxylin-eosin, x 50).



Fig. 6. Lymphangioma (hematoxylin-eosin, x 400).

non-homogenous echogenicity and endophytic growth pattern. The uterus and the right adnexa were unremarkable. There was no free fluid in the pouch of Douglas. Contrast-enhanced computed tomography (CT) of the abdomen and pelvis showed a cystic lesion with segmental



**Fig. 5.** Lymphangioma (CD31, x 100).



Fig. 7. Lymphangioma (podoplanin, x 50).

wall thickening, measuring 133 x 83 x 135 mm, adjacent to the left ovary and the uterus on the left side. The lower part of the lesion displayed a non-homogenous hyperechoic pattern of segmental wall thickening up to 15 mm and ca. 42 mm in length which could correspond to the involved left ovary. The lesion, localized by means of CT on the left side, displaced the small intestine mesentery in a upward direction and to the right. The right ovary measured 43 x 24 x 46 mm and had a multicystic structure. Colonoscopy showed first-degree hemorrhoids, but the findings were otherwise unremarkable.

After bowel preparation, laparotomy was performed using a Pfannenstiel incision. When the peritoneal cavity was entered the presence of massive adhesions was noted between the uterus and the small intestine, large intestine and omentum. The right adnexa was adherent to the vermiform appendix and omentum. The left ovary cyst, measuring ca. 60 mm, was adherent to the left fallopian tube. Above the uterus, on the left side, in the area between the lower and middle abdomen, a giant cyst, ca. 20 cm in diameter was exposed. It was greenish-black in color which suggested necrosis and its exact point of origin was difficult to identify. Adhesions blocked access to the pouch of Douglas. The parietal peritoneum, urinary bladder area and omentum were not involved. Since the access to the cyst was difficult, the incision was extended longitudinally up to 5 cm above the umbilicus and intraoperative abdominal surgical consultation was requested. The giant cyst was located in the mesentery at the junction of the small and large intestine. A stricture of the sigmoid colon was seen at the rectosigmoid junction. The adhesions between the loops of the intestine and the cyst were released by sharp and blunt dissection, the cyst ruptured during the procedure and its capsule was excised. Standard appendectomy was performed followed by amputation of the uterus with the left adnexa after previous release of adhesions. The right adnexa was densely adherent to the omentum and macroscopically non-suspicious, and it was decided not to proceed with their excision. The intraoperative blood loss was estimated at 700 mL. The cyst fluid was sent for cytology and culture and the tissue specimens for histological examination. Histopathological examination of the uterine specimens showed several leiomyomas up to 1 cm in diameter. Foci of endometriosis were found in the parametria, left ovary and fallopian tube, and vermiform appendix. Lymphangioma was diagnosed in the postoperative specimens of the small and large intestine mesentery. The cyst fluid cultures were negative for bacteria and fungi as was the cytology for malignancy.

## DISCUSSION

Lymphangiomas usually occur in the mesentery of the small or large intestine and in most cases are asymptomatic. In this case, lymphangioma was found in an even less common location, in the mesentery of the ileocecal valve, but it did not produce any of the signs and symptoms described in the literature such as intestinal obstruction, constipation, vomiting [2], abdominal distension consistent with the lymphangioma size [4] or gradually increasing non-specific abdominal pain persisting for a few days to a few years [5-10]. Lymphangiomas are very difficult if not impossible to diagnose without a definitive histopathological diagnosis. There have been attempts to puncture cystic lesions of the mesentery and perform the cytologic examination of the

aspirated fluid, but findings were inconclusive. Computed tomography (CT) is recognized as the basic imaging study for the differential diagnosis of cysts in the mesentery and extraperitoneal space, other than lymphangioma [2-11]. In cases reported in the literature, CT was used to establish the site of origin of the cysts, choose the appropriate operative technique [5, 7, 8] and the extent of the procedure. CT is also used to observe the lesion growth and as they expand the involvement of other organs [7, 9].

Pre-operative CT was also used in the present case but it failed to establish the exact point of origin of the cyst. The CT report suggested the left ovary as the site of origin of a malignant lesion and advised proceeding to magnetic resonance imaging (MRI). According to the CT report, the lesion displaced the mesentery in an upward direction, which suggested that there was no direct connection between the lesion and the mesentery. If the CT had showed a cyst in the mesentery, a different surgical approach would have been chosen. A longitudinal incision extending from the umbilicus to the symphysis pubis would have been used, instead of a Pfannenstiel incision which does not provide a good approach in bowel surgery.

On the other hand, abdominal ultrasound and, depending on the lesion location, transvaginal ultrasound allow initial identification of any abnormal structure and the findings dictate further diagnostic workup and appropriate management in cases of suspected cystic lesions of the mesentery. Since the signs and symptoms of mesenteric lymphangioma are often absent or appear late, in women a regular gynecological check-up consisting of a bimanual examination, a cervical cytology test and transvaginal ultrasound may be the first step leading to further diagnostic procedures in this rare but serious condition.

In cases of any doubts concerning the exact location of the lesion, we recommend using magnetic resonance imaging (MRI) as it adds value to the diagnostic workup. It helps to identify the point of origin of the lesion and so would aid in selecting the appropriate surgical technique and if required including other specialists in the surgical team. Also, when the exact location of the lesion is unclear, prior to surgery, members of the team should discuss in detail the planned procedure. The nature of the surgery must be explained to the patient who should be warned of any possible extensions. In the reported case, the patient was informed that the procedure could involve not only the removal of the left ovary and its lesion, but also hysterectomy and right adnexectomy. If it were not possible to extirpate the lesion in its entirety from the ileocecal region, creation of a temporary stoma would be likely with a reversal in the future. The surgeon must provide the patient with information about all possible scenarios and the patient must confirm that she fully understands the procedure by signing the consent for surgery form.

#### CONCLUSIONS

1. We recommend using magnetic resonance imaging (MRI) to determine the exact location of the lesion.

- 2. MRI helps to identify the point of origin of the lesion and select the appropriate surgical technique in many cases.
- 3. Members of the surgical team should always discuss the details before the planned operation.
- 4. In case of possible complications during the surgery, patient should be warned in advance about alternative procedures.

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# **Conflict of interest:**

The Authors declare no conflict of interest.

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Received: 08.03.2022 Accepted: 27.08.2022

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- $\mathbf{D}$  Writing the article,  $\mathbf{E}$  Critical review,  $\mathbf{F}$  Final approval of the article



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