SERVİKSİN KAVERNÖZ LENFANJİOMASI

Cavernous Lymphangioma of the Uterine Cervix

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ÖZET

Lenfanjiyomlar lenfatik sistemin nadir görülen benign proliferasyonlarıdır. Genel olarak üç tip kabul edilmiştir: sirkumskriptum (ya da kapiller), kavernöz ve kistik. Biz 54 yaşındaki kadında serviksde kavernöz lenfanjioma olgusunu sunuyoruz. 54 yaşındaki kadın kadın hastalıkları ve doğum kliniğine başvurdu. Uterine myoma nedeniyle, total abdominal histerektomi ve bilateral salpingo-ooferektomi uygulandı. Ameliyat materyali patoloji bölümüne gönderildi. Makroskopik değerlendirmede, serviks kesit yüzeyinde çok sayıda siyahımsı renkte kavitasyonlar ortaya çıktı. Mikroskobik incelemede, lenfatik sıvı içeren kavernöz lenfatik alanlardan oluşan kavernöz lenfanjiom gözlendi. İmmünhistokimyasal olarak, faktör VIII, CD34, SMA, D2-40 lenfatik endotelinde pozitifti. Kavernöz lenfanjiyom, kadınlarda serviksde görülmesi nadir bir lezyondur. Bizim olgumuzda olduğu gibi, serviks ya da kadın genital sistemde başka bir bölgenin vasküler lezyonları histerektomi materyallerinde tesadüfen tespit edilebilir.

Anahtar Sözcükler: *Lenfanjioma; Uterine serviks; Genital trakt.*

ABSTRACT

Lymphangiomas are rare benign proliferations of the lymphatic system. Three types are generally acknowledged: circumscriptum (or capillary), cavernous, and cystic. We present a case of cavernous lymphangioma of the cervix in a 54-years-old female. A 54-years-old woman presented to the department of obstetrics and gynecology. For uterine myoma, she underwent total abdominal histerectomy and bilateral salpingo-oophorectomy. Operation material was sent to pathology department. Macroscopic evaluation revealed, cut surface of cervix was blackish and had many cavities. Microscopic examination showed cavernous lymphangioma; composed of cavernous lymphatic spaces which contain lymphatic fluid. Immunhistochemically, factor VIII, CD34, SMA, D2-40 were positive in lymphatic endothelium. Cavernous lymphangioma is a rare lesion in the cervix of females. To our knowledge, this is one of the few cases documented in literature. As our case, vascular lesions of cervix or another site of female genital tract can be detected coincidentally at histerectomy materials.

Keywords: *Lymphangioma; Uterine cervix; Genital tract.*

INTRODUCTION

Lymphangiomas are rare benign proliferations of the lymphatic system. These malformations can occur at any age and may involve any part of the body, but 90% occur in children less than 2 years of age and involve the head and neck. These malformations are either congenital or acquired. Lymphangiomas have traditionally been classified into three subtypes: capillary (circumscriptum), cavernous and cystic.

Cavernous lymphangiomas are composed of dilated lymphatic channels, characteristically invade surrounding tissues.

Cavernous lymphangioma is an uncommon lesion in the uterine cervix of females. Only a few cases have been documented in literature.

We present a case of cavernous lymphangioma of the cervix in a 54 years old female.

CASE REPORT

A 54-year-old woman had admitted to gynecology outpatient clinics of Abant Izzet Baysal University Hospital. She had an obstetric history of 3 gestations, 2 resulted with birth, 1 with ectopic pregnancy. Also she had medical history of asthma, hypertension, cholecystectomy and ectopic pregnancy operations. She had complaints frequent urination. Pelvic examination revealed an intramural myoma and surgery was planned. Tumor markers were within normal range. The patient had undergone total abdominal hysterectomy and bilateral salpingo-oophorectomy operation.

At macroscopic examination uterus was 105x65x55 mm in dimensions and 191 grams in weight. In the same container, with the uterus, there was myomectomy material which was 11x8x8 cm in dimensions and 375 grams in weight.

Cut surface of cervix had milimetric cavities and its colour was blackish.

In microscopic examination sections of the cervix showed cavernous lymphangioma composed of cavernous lymphatic spaces which contain lymphatic fluid (Figure 1-2).

Figure 1: Endoservical glands and lymphatic spaces (H&E, X40).

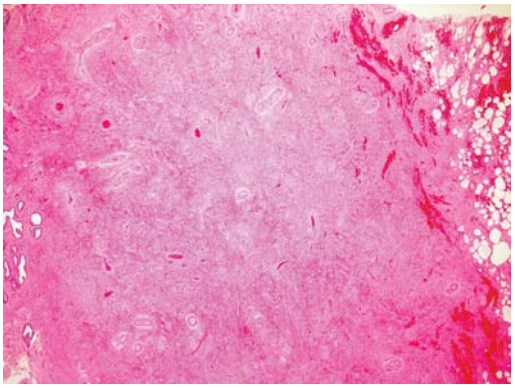
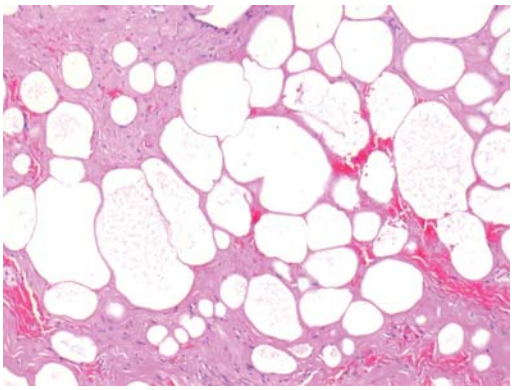


Figure 2: Cavernous lymphatic spaces (H&E, X100).



Immunohistochemically, Factor VIII, CD34, SMA and D2-40 were positive in lymphatic endothelium (Figure 3-5).

Figure 3: SMA+ lymphatic endothelium (X200).

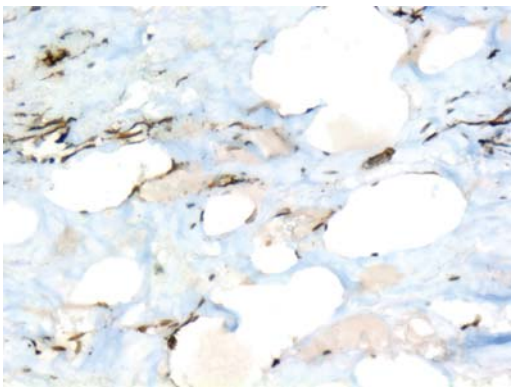


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Figure 4: D2-40+ lymphatic endothelium (X200).

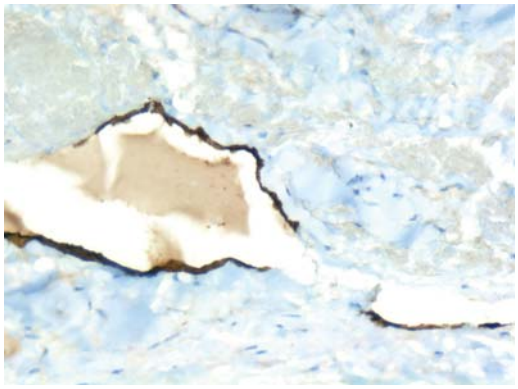
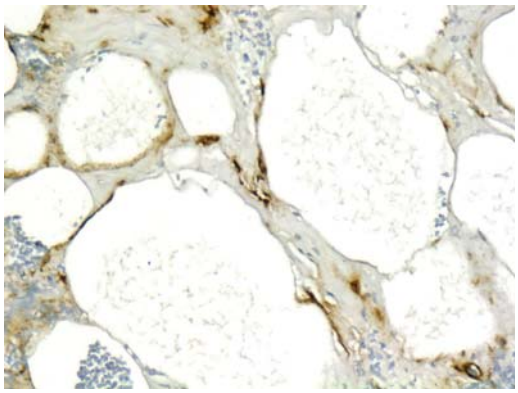


Figure 5: CD34+ lymphatic endothelium (X200).



DISCUSSION

Lymphangiomas are relatively uncommon lesions, more so in the uterine cervix, where they are very rare. Some authorities regard lymphangiomas to be true neoplasms that are capable of local agressive behavior, but overall they are benign (1).

By contrast, others believe that lymphangiomas are hamartomatous malformations that result from the failure of the lymphatic system to communicate with the venous system. They may result from trauma, inflammation or lymphatic obstruction. Another closely related view is that lymphangiomas represent sequestrated lymphatic tissue that fails to communicate normally with the lymphatic system. According to these latter two hypotheses, abnormal rests of lymphatic tissue possess some capacity to proliferate and accumulate vast quantities of fluid, accounting for their cystic appearance (1).

It may even be that, our case had a lymphangioma of the cervix caused by previous surgery, myoma or chronic inflammation.

Histologically, cavernous lymphangiomas show numerous narrow spaces containing amorphous eosinophilic fluid and few lymphocytes and lined by a monolayer of attenuated endothelial cells. Some of the channels may contain blood because of secondary haemorrhage, resulting in misdiagnosis of this lesion as cavernous hemangioma. However, the presence of large collections of lymphoid cells in the stroma, sometimes with lymphoid follicle formation, and the relatively greater irregularity of the lumina of the cavernous spaces, tilt the diagnosis in favour of lymphangioma. Vascular endothelial markers such as factor VIII- associated antigen, CD 31, CD 34, SMA may be positive in lymphatic endothelium (1,2).

Andola Uma S and Andola Sainath K, in 10 years period, reported 11 vascular tumors of the female genital tract which all were benign vascular neoplasms. The most common lesion was the hemangioma (five). Other benign lesions included lymphangioma (two), lymphangioma circumscriptum (one), chorangioma (one), angiomyofibroblastoma (one), angiomyxoma (one). The vascular tumors occured most commonly in ovary (five), followed by vulva (three), and one each in cervix, vagina and placenta (3).

In another study, 10 patients with benign vascular tumors of female genital tract were evaluated over a 4 years period. The presenting complaint was abdominal pain/mass, postcoital bleeding, and vaginal and vulval mass. Of these, histologically, all were benign vascular neoplasms, ranging from hemangioma (five), lymphangioma (one), lymphangioma circumscriptum (one) to angiomatosis (two) and arteriovenous malformation (one). The vascular tumors arised most commonly in ovary (six), followed by vulva (two) and one each in cervix and vagina (4).

Gerbie AB et al, in 1955, found only 1 lymphangioma of the cervix in the literature. A huge hyperthropic growth of the cervix developed during pregnancy. Microscopically it was proven to be a lymphangioma (5).

In a review of 15000 specimens of the female genital system was examined during 10 years, nine cases of vascular tumors were found (capillary hemangioma of the endometrium, capillary and cavernous hemangioma of the cervix and vulva, angiokeratoma of the vulva, hemangiosarcoma of the mons pubis) by Kondi-Pafiti et al. Cavernous lymphangioma wasn't identified (6).

In conclusion, cavernous lymphangioma is a rare lesion in the uterine cervix. As our case, some are asymptomatic and are found incidentally. Vascular tumors especially of the cervix, can be difficult to determine clinically and radiologically. A pathological examination is necessary in all such cases. Surgical excision is curative in most of the cases.

"Conflict of interest: None"

REFERENCES

1. Weiss SW, Goldblum JR: Tumour of Lymph vessels. In Enzinger and Weiss's Soft tissue tumours 4th edition. St Louis: Mosby; 2001: p 955-98.
2. Rosai J: Soft tissues. In Rosai and Ackerman's Surgical Pathology Volume 2. 9th edition. St. Louis: Mosby; 2004: p 2237-71.
3. Andola Uma S, Andola Sainath K. Vascular Tumours of the Female Genital Tract: A Clinicopathologic Study of 11 Cases. Journal of Clinical and Diagnostic Research. 2011; 5(6): 1241-6.
4. Gupta R, Singh S, Nigam S, Khurana N. Benign vascular tumors of female genital tract. Int J Gynecol Cancer. 2006; 16(3): 1195-200.
5. Gerbie AB, Hirsch MR, Greene RR. Vascular tumors of the Female Genital Tract. 1955; 6(5): 499-507.
6. Kondi-Pafiti A, Kairi-Vassilatou E, Spanidou-Carvouni H, Kontogianni K, Dimopoulou K, Goula K. Vascular tumors of the Female Genital Tract: A Clinicopathological Study of Nine Cases. Eur J Gynaecol Oncol. 2003; 24(1): 48-50.