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CLINICAL CASE

Lymphangioma of the ovary: a case report and review of the literature

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Summary

Lymphangioma of the ovary is a very rare tumor which is usually silent and identified incidentally at operation or autopsy. We report on a 61-year-old woman with lymphangioma of the ovary and review the relevant literature. The microscopic examination of the left ovary revealed numerous vascular spaces of different dimensions of which the inner surfaces were lined with flattened endothelial cells with neither cellular atypia nor extraluminal or intraluminal proliferation. There was no evidence of haemorrhage or necrosis. The stroma was formed by fibrocollagenous tissue infiltrated by rare lymphocytes. The cells lining vascular spaces were immunoreactive for CD31 by immunohistochemical staining.

Key words: lymphangioma, ovary, tumor

Introduction

Lymphangioma of the ovary is a very rare tumor which is usually silent and identified incidentally at operation or autopsy. Kroemer first described this tumor in 1908; in his description emphasised the features of the disease including cystic spaces that are lined by endothelial cells and contain serous or chylous fluid [1].

We report on a 61-year-old woman with lymphangioma of the ovary and review the relevant literature.

Case presentation

A 61-year-old menopausal woman was admitted at Cerrahpasa Medicine Faculty, Department of Gynaecology with lower abdominal pain. There was no relevant prior history. On physical examination an abdominal mass was palpable. Abdominopelvic ultrasound demonstrated a large, cystic and solid mass filling the abdominal cavity.

The patient underwent abdominal hysterectomy with bilateral salpingo-oophorectomy. Frozen section of the resected organs showed two distinct tumors; one was an uterine leiomyoma with cystic degeneration which, on gross pathologic examination, was 35 cm in diameter and located in the posterior wall of the uterus, while the left ovary had a tumor which measured approximately 9×3×0.5 cm, the cut surface of which showed cysts with a maximum dimension of 1 cm (Figure 1).

The microscopic examination of the left ovary revealed numerous vascular spaces of different dimensions of which the inner surfaces were lined with flattened endothelial cells with neither cellular atypia nor extraluminal or intraluminal proliferation. There was no evidence of haemorrhage or necrosis. The stroma was formed by fibrocollagenous tissue infiltrated by rare lymphocytes (Figure 2). The cells lining vascular spaces were immunoreactive for CD31 by immunohistochemical staining (Figure 3). Corpus albicans were seen in the surrounding ovarian tissue.

In addition, microscopic examination of the uterus and fallopian tubes showed chronic nonspesific cervicitis, an endometrial polyp, an uterine leiomyoma and numerous paratubal cysts.

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Figure 1. Numerous ovarian cysts on the cut surfaces.

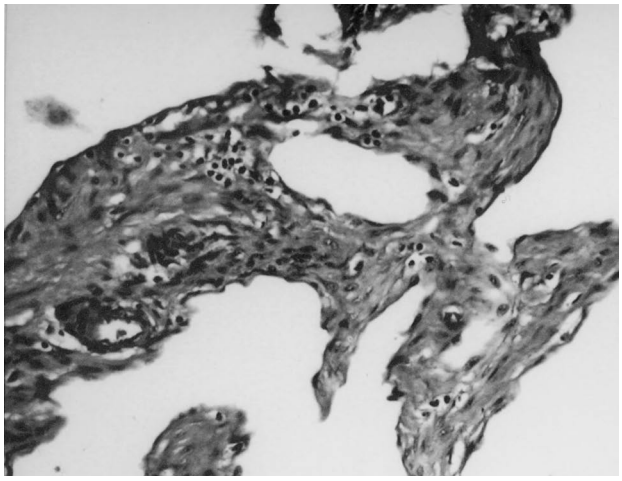

Figure 2. Numerous vascular spaces lined with flattened endothelial cells with neither cellular atypia nor extraluminal or intraluminal proliferation (H&E ×100).

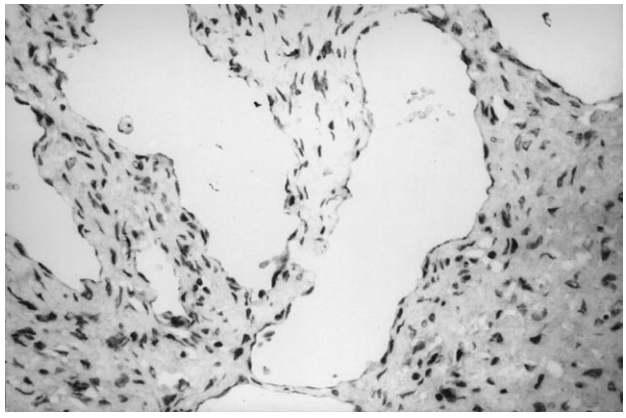

Figure 3. The cells lining vascular spaces are immunoreactive for CD31 by immunohistochemical staining (×100).

Discussion

Lymphangioma of the ovary is a rare lesion [2]. Around 16 cases have so far been reported in the literature and in only 3 of these cases bilateral ovarian involvement has been described [1,3].

Ovarian lymphangioma is often asymptomatic and is identified incidentally at operation or autopsy or as a result of medical tests [4]. Ahluwalia et al. [4] reported a case with an ovarian lymphangioma in the right ovary 6×6×4 cm in diameter, found incidentally at abdominal ultrasound for cholelithiasis. Our case presented with lower abdominal pain caused by the huge uterine leiomyoma.

The pathogenesis of ovarian lymphangioma is still unclear. Some authors have suggested that they are true neoplasms whilst others consider them to be the result of lymphatic proliferation caused by hamartoma, malformations, adenomatoid tumors, lymphatic circulation disorders or scarred regional lymph nodes [1,5]. Evans et al. [1] reported bilateral ovarian lymphangiomas in a patient with bilateral chronic follicular salpingitis and suggested they were the result of a lymphatic circulation disorder. However, in one of the other reported cases with ovarian lymphangioma which was detected during an examination for cholelithiasis, there was no evidence of salpingitis [4]. In one case a patient with ovarian lymphangioma had undergone nephrectomy and had also received radiation therapy and chemotherapy for Wilms' tumor when she was 6 years old, so this case was attributed to a lymphatic drainage disorder [3]. In our case, we, therefore, attribute the lymphangioma of the ovary to lymphatic circulation disorder caused by the leiomyoma of 35 cm in diameter.

Some authors have suggested that the lesion is always benign and curable by resection. There are, however, 2 reported cases with relapse in the literature. Rice et al. [6] reported a case with metastasis in the right ovary, peritoneum and liver parenchyma 6 months after resection of the lymphangioma in the left ovary. Another recurrent lymphangioma of the ovary was described by Aristizabal et al. [7], which recurred in the peritoneal cavity 2 years after the operation.

Because the malignant potential of this lesion is still unknown, long-term and careful follow-up should be considered in these patients. Our patient has remained healthy for 14 months after the operation.

References

1. Evans A, Lytwyn A, Urbach G, Chapman W. Bilateral lymphangiomas of the ovary: an immunohistochemical

characterization and review of the literature. Int J Gynecol Pathol 1999; 18: 87-90.
2. Prat J, Fox H. Mesenchymal tumors of the ovary. In: Fox H, Wells M (ed): Obstetrical and Gynaecological Pathology. New York, Churchill Livingstone, 1995 (5th edn), p 870.
3. Heinig J, Beckmann V, Bialas T, Diallo R. Lymphangioma of the ovary after radiation due to Wilms' tumor in the childhood. Eur J Obstet Gynecol Reprod Biol 2002; 103: 191-194.
4. Ahluwalia J, Girish V, Saha S, Dey P. Lymphangioma of the ovary. Acta Obstet Gynecol Scand 2000; 79: 894-895.
5. Talerman A. Nonspecific tumors of the ovary. In: Kurman RJ (ed): Blaustein's Pathology of the Female Genital Tract. New York , Springer Werlag, 2002 (5th edn), p 1044.
6. Rice M, Pearson B, Treadwell WB. Malignant lymphangioma of the ovary. Am J Obstet Gynecol 1943; 45: 884-888.
7. Aristazabal SA, Galindo JH, Davis JR, Boone ML. Lymphangiomas involving the ovary. Report of a case and review of the literature. Lymphology 1977; 10: 219-223.