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Leiomyosarcoma of the Broad Ligament: A Case Report and Literature Review

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A 58 year old Bahraini with leiomyosarcoma of the broad ligament (LBL) is reported. Treatment consisted of extensive dissection of tumor, omentectomy, total abdominal hysterectomy, bilateral salpingo-oopherectomy and the removal of pelvic and abdominal wall metastases. Post-operatively the patient was treated with radiotherapy.

This is the first reported case of LBL from the Arabian Gulf region and the eleventh case in the world literature. It has been described here not solely because of its rarity, but to highlight the challenges associated with its management.

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The broad ligament variety of leiomyosarcoma is extremely rare, and only 10 cases have been reported so far in the medical literature. We present this case because of the nature of LBL and its management which has been scarcely reported and we aim at adding to the clinical experience about the management of this tumour. A review of the medical literature will also be presented.

THE CASE

A 58 year old postmenopausal woman, Gravida 11, Para 5, weighing 80 Kgs, was admitted to hospital with a history of low-grade pyrexia, loss of weight and anorexia for 4 months. On admission, she complained of persistent nausea, vomiting and a gross painless abdominal swelling. She had no alteration in bowel or bladder habits. She was known to have hypertension and type II diabetes and was on regular treatment. She had no previous surgical intervention. Menopause had occurred four years earlier, and she had had no postmenopausal bleeding or vaginal discharge since that time.

On abdominal examination, a large, solid tender and non-mobile mass with an irregular outline was felt. It was equal in size to a 20-week pregnancy. The mass was arising from the pelvis and felt more appreciably towards the left side. It extended towards the left lumbar and hypochondrial regions. There was no evidence of regional lymphadenopathy, and no sign of free fluid in the peritoneum. The liver and kidneys were not palpable.

Speculum vaginal examination showed atrophic changes. On bimanual examination, the uterus could not be distinguished from the mass. The pouch of Douglas did not feel nodular. Rectal examination revealed no abnormality.

Ultrasonography confirmed the presence of an hyperechoic, solid mass, with a diameter of 18 x 12 x 18 cm arising from the left side of the pelvis.

Laboratory findings were all within normal limits including tumour markers CA-125, Alpha-feto protein and Beta-HCG. A cytology smear was performed. Results were normal.

Enhanced contrast CT scans for kidneys, liver, spleen and pancreas was normal, and reported as follows. "There was no hydronephrosis or lymphadenopathy in the retroperitoneum. A very large well defined solid, soft tissue mass was seen arising from the pelvis and reaching superiorly up to the uterus, indenting the bladder downwards and extending to the anterior abdominal wall".

At Laparotomy the uterus was slightly enlarged. The tubes and ovaries were normal. The left adnexal mass was approximately 14 x 16 cm, with fungating growth of friable gelatinous material on the surface. It extended towards the left lumbar area, the urinary bladder and the lower part of the anterior abdominal wall. The mass was firm and immobile. It engulfed the uterus anteriorly and posteriorly and indented the bladder. Multiple metastases were found in the pelvic peritoneum. Liver, spleen and kidneys were felt and found to be normal. There was no ascies or gross lymphadenopathy. The report of a frozen section showed cellular leiomyosarcoma. Dissection of the tumour combined with total abdominal hysterectomy and bilateral salpingo-oopherectomy and omentectomy were performed. Samples

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of pelvic lymph nodes were obtained and sent for histology.

Convalescence was rapid apart from Pseudomonas urinary tract infection, which responded to a course of Aztreonam (Azactam) injections for 6 days. The patient was discharged on the 12th postoperative day.

Pathological description: The specimen included uterus, both ovaries and tubes and a large lobulated tumour 12 cm in diameter occupying the left broad ligament area. The left tube is stretched over the tumour and the left ovary was clearly separate. The tumour was capsulated and a plane of cleavage between the mass and uterus was present. Cut slices showed an enlarged body of uterus and cervix, and the tumour, separate from the uterus. The specimen showed a variegated texture with hemorrhage, yellowish necrotic and fleshy myxomatous areas all over (Fig 1 and 2). A benign endometrial polyp and small intramural leiomyoma were seen. Both tubes and ovaries were unremarkable and the omentum was free of tumour.

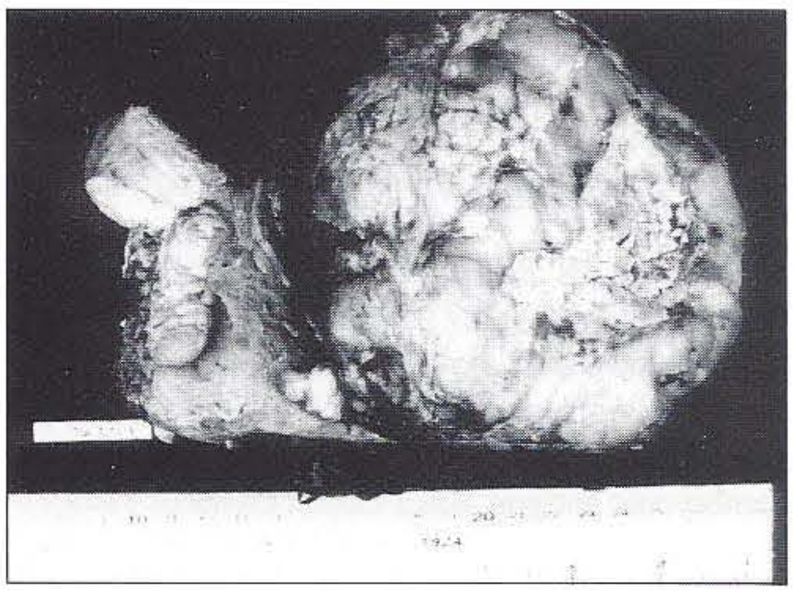

Figure 1. Specimen of uterus (right side) with large lobulated and capsulated tumour in the broad ligament. The two ovaries (OV), fallopian tube (FT) and cervix (CX) are shown.

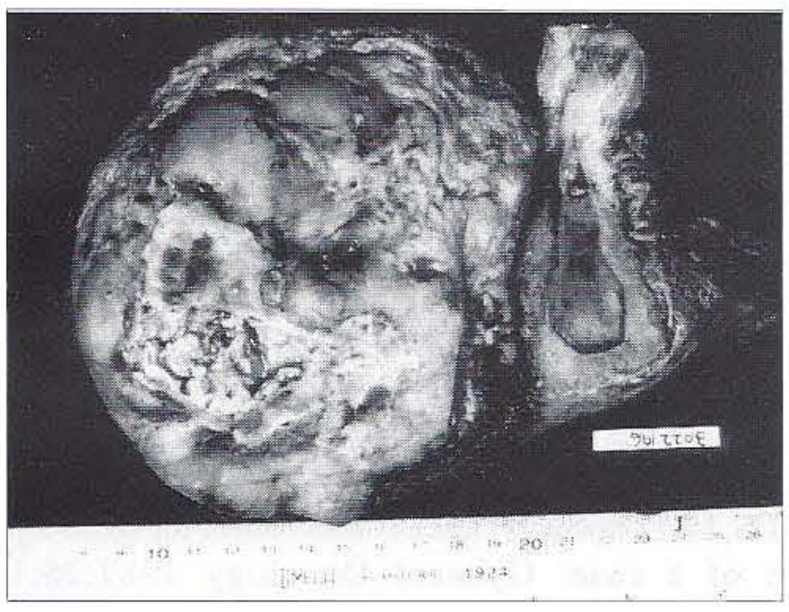

Figure 2. Cut surface of uterus and tumour. The plane of cleavage between the uterus and the tumour is clearly seen. The tumour cut surface show necrosis and myxomatous nodules.

Histology: Figure 3 and 4 shows the highly pleomorphic spindle cell structure with bizarre cells. There were large areas of necrosis, oedema and myxomatous change. Mitotic figure count varied up to as many as 8 figures in one high power field. The rest of the organs including: cervix, endometrium, myometrium and both ovaries and tubes were unremarkable.

A diagnosis of leiomyosarcoma of left broad ligament was made. Histological grade was moderate. No involvement of uterus was detected.

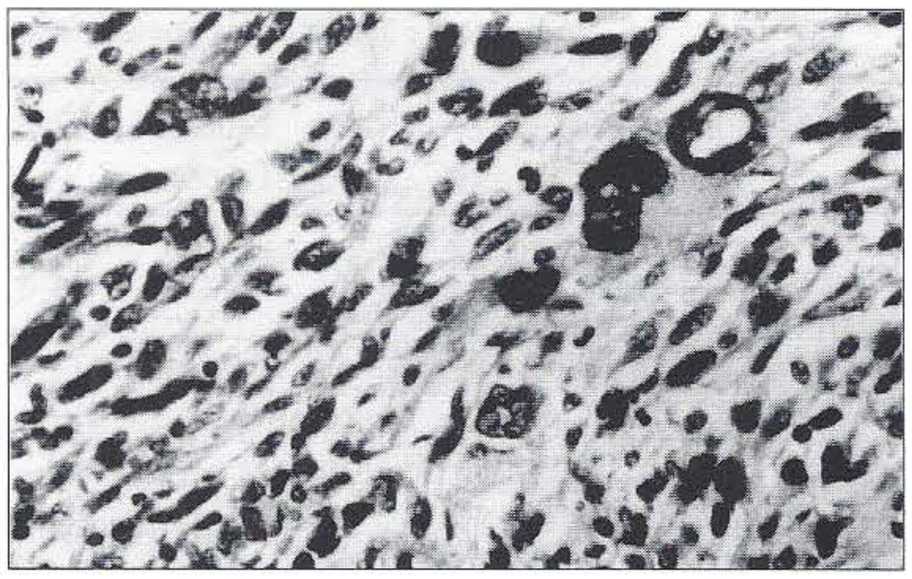

Figure 3. Photomicrograph of tumour to show a leiomyosarcoma with pleomorphism of size and shape of cells (H&E x 40).

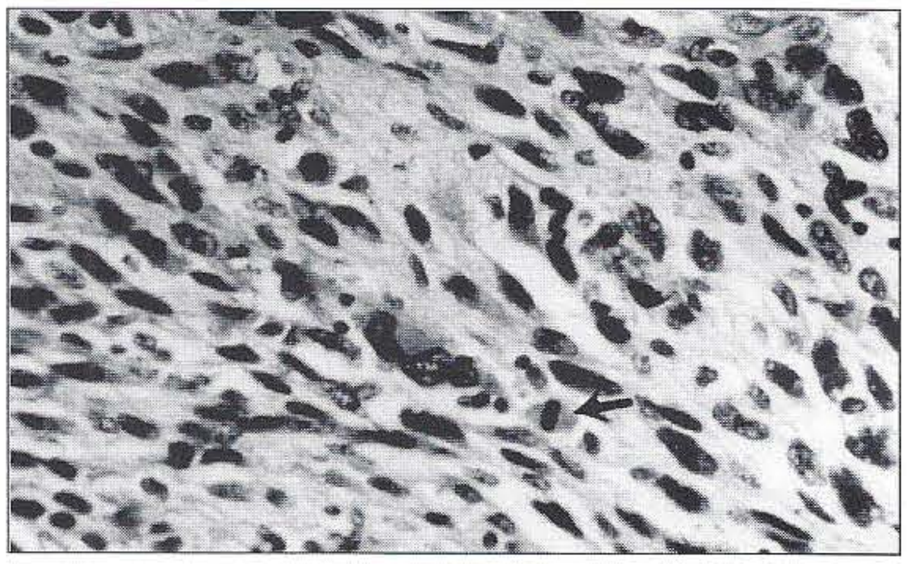

Figure 4. Photomicrograph of leiomyosarcoma showing the pleomorphism. Three mitotic figures are shown (arrows).

Follow-up: At the 4th postoperative week she was seen by an oncologist who suggested CT scan as a base line study following surgery. The report was as follows: "Matted pelvic bowel loops. Soft tissue mass". Seven weeks postoperatively CT scan was repeated which revealed recurrence of tumour 12 x 10 cm superior to the bladder.

In review of the above finding, it was suggested that the patient have palliative radiotherapy.

DISCUSSION

Only ten similar cases have been reported in the literature, from 1960 to 1998. This is the first case of leiomyomasarcoma of the broad ligament from the Gulf States.

In the early part of this century German and French authors have described several cases of sarcomas at this site[9]. Most of these reports, however, did not adhere to the criteria of definition given by Gardner et al, who emphasized that these tumours must occur in or on the broad ligament, but are separate from and in no way connected with either the uterus or the ovary. The case reported here is compatible with these criteria. See plate no. (1 and 2).

The clinical manifestation of most of the previously reported cases was non-specific including abdominal pain, abdominal distention, constipation, nausea and malaise. Occasionally there were progressive symptoms or an acute retention of urine and no case was diagnosed before the operation[3,4].

In most reports, the cases described were in 6th decade. Ullmann and Roaumell described two women in 1973, and in 1995 one by Cheng et al[5,6]. The latter patient was admitted to hospital with the lesion well advanced, and despite

Table 1: Similar reported cases of Leiomyosarcoma from 1960–1998

Case	Author/Ref No.	Age	Side of Tumour	Size (cm)	Mitoses HPF	Initial treatment	Subsequent treatment	Survival
1	Lowell and Karsh (5)	50	Right	11 x 7.5	0-4	TAH, BSO	–	>12 months
2	Ullmann and Roumell (6)	50	Left	11 x 7.5 x 6.5	15	TAH, BSO	RT, CT	7 months, DOD
3	Weed and Podger (7)	50	Left	11 x 3 x 7	12	TAH, BSO	CT	19 months, DOD
4	DiDomenico et al(8)	48	Left	11 x 10 x 9	10.5	TAH, BSO	–	No report
5	Raj-Kumar (9)	70	Left	10 x 7 x 6.5	10	Enucleation	–	No report
6	Herbold et al (10)	73	Left	15	21	TAH, BSO	–	1 month, DOD
7	Shimm and McDonough (11)	31	Right	9 x 7 x 4	8	Enucleation, RT	CT	>30 months
8	Lee et al (12)	36	Left	35 x 30 x 5	>10	TAH, BSO, CT	RT	>33 months
9	Lee et al	65	Left	16.5 x 12.6 x 12.4	>10	Subtotal Hysterectomy, BSO, CT	–	30 months, DOD
10	Cheng et al (1)	59	Right	7 x 6 x 6	>10	TAH, BSO	–	Alive >12 months

TAH, BSO - Total Abdominal Hysterectomy and Bilateral Salpingo Oopherectomy

CT – Chemotherapy RT – Radiotherapy DOD – Died of Disease

extensive surgery and radio-therapy she was readmitted six months later with recurrences, but she died at the eighth month postoperatively. Gross survival rate quoted for all leiomyosarcoma is 8-46%. Previous reports stated a crude survival rate of 75% in-patients who were premenopausal at the time of diagnosis and 37% in postmenopausal women[4].

From a histopathological point of view it is important to differentiate between atypical varieties such as smooth muscle leiomyoma and benign metastasizing leiomyoma, from leiomyosarcoma. The criteria for diagnosing malignancy in these cases are: more than eight mitosis per high power field (hpf), based on counts of 40 or more per hpf in the most cellular areas. However, tumours or extensions beyond the uterus at the time of diagnosis are considered malignant regardless of mitosis count and degree of pleomorphism, although there can be wide variations in the degree of aggressiveness (Fig 3 & 4).

With regard to postoperative follow-up, Parente[3] has reported favourably on the use of carcinoembryonic levels, in assessing both the degree of malignancy and the response of the leiomyosarcoma to surgical removal of the tumour, chemo and radiotherapy.

Treatment with chemotherapeutic agents, radiotherapy alone or combined with surgery, appears to have only variable benefit[4].

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