Comparison of retroperitoneal liposarcoma extending into the inguinal canal and inguinoscrotal liposarcoma

Jinsoo Rhu, MD
Chan Woo Cho, MD
Kyo Won Lee, MD
Hyojun Park, MD, PhD
Jae Berm Park, MD, PhD
Yoon–La Choi, MD, PhD
Sung Joo Kim, MD, PhD

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Correspondence to:
S.J. Kim
Department of Surgery
Samsung Medical Center
Sungkyunkwan University School of Medicine
50 Irwon-dong, Gangnam-gu
Seoul 135-710, Korea
kmhyj.kim@samsung.com

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Background: This study was designed to analyze differences between retroperitoneal liposarcoma (RLPS) extending into the inguinal canal and inguinoscrotal liposarcoma.

Methods: We retrospectively reviewed the records for patients who were managed for inguinal liposarcoma at Samsung Medical Center, a tertiary hospital, between January 1998 and December 2016. Patient data on demographics, tumour location, surgery, adjuvant therapy, histology, recurrence and death were collected. We used Mann–Whitney, Fisher exact and Kaplan–Meier log-rank tests to analyze differences between groups.

Results: Seven of 179 (3.9%) patients with abdominal liposarcoma had inguinoscrotal liposarcoma, and 6 of 168 (3.6%) patients with RLPS had extension to the inguinal canal. No differences were observed between groups in sex ($p > 0.99$), mean age (49.7 ± 6.4 yr v. 52.1 ± 12.5 yr, $p = 0.37$), laterality ($p > 0.99$) or scrotal involvement (40.0% v. 66.7%, $p = 0.57$). The RLPS group had significantly larger tumours than the inguinoscrotal group (27.9 ± 6.8 cm v. 7.8 ± 4.2 cm, $p = 0.001$). Postoperative complications were significantly more common in the RLPS group ($n = 4$, 83.3%); patients in the inguinoscrotal group experienced no postoperative complications ($p = 0.021$). Log-rank tests showed that the groups had no statistical differences in disease-free survival ($p = 0.94$) or overall survival ($p = 0.10$). However, inoperable disease-free survival was significantly poorer in the RLPS group ($p = 0.010$).

Conclusion: Although initial signs and symptoms can be similar, RLPS extending into the inguinal canal was associated with significantly higher morbidity and mortality than inguinoscrotal liposarcoma.

Contexte : Cette étude visait à examiner les différences entre le liposarcome rétropéritonéal s'étendant au canal inguinal et le liposarcome inguino-scrotal.

Méthodes : Nous avons procédé à une analyse rétrospective des dossiers de patients traités pour un liposarcome inguinal au Samsung Medical Center, un hôpital de soins tertiaires, entre janvier 1998 et décembre 2016. Nous avons recueilli les données des patients en ce qui a trait aux caractéristiques démographiques, au siège de la tumeur, à la chirurgie, au traitement adjuvant, à l'histologie, à la récidive et au décès. Nous avons utilisé le test de Mann–Whitney, la méthode exacte de Fisher et les tests logarithmiques par rangs de Kaplan–Meier pour analyser les différences entre les 2 groupes.

Résultats : Sept des 179 (3,9 %) patients atteints de liposarcome abdominal avaient un liposarcome inguino-scrotal, et 6 des 168 (3,6 %) patients atteints de liposarcome rétropéritonéal présentaient une extension au canal inguinal. Aucune différence n'a été observée entre les groupes pour le sexe ($p > 0,99$), l'âge moyen (49,7 ± 6,4 ans c. 52,1 ± 12,5 ans, $p = 0,37$), la latéralité ($p > 0,99$) ou l'atteinte scrotale (40 % c. 66,7 %, $p = 0,57$). La taille de la tumeur était significativement plus grande dans le groupe du liposarcome rétropéritonéal que dans celui du liposarcome inguino-scrotal (27,9 ± 6,8 cm c. 7,8 ± 4,2 cm, $p = 0,001$). De même, les complications postopératoires étaient significativement plus courantes dans le groupe du liposarcome rétropéritonéal ($n = 4$, 83,3 %), les patients du groupe du liposarcome inguino-scrotal n'en ayant pratiquement pas présenté ($p = 0,021$). Les tests logarithmiques par rangs ont révélé l'absence de différences statistiques entre les groupes pour la survie sans récidive ($p = 0,94$) et la survie globale ($p = 0,10$). Cependant, la survie sans récidive du patient inopérable était significativement plus faible dans le groupe du liposarcome rétropéritonéal ($p = 0,010$).

Conclusion : Malgré la similarité des premiers signes et symptômes, le liposarcome rétropéritonéal s'étendant au canal inguinal était associé à des taux de morbidité et de mortalité significativement plus élevés que le liposarcome inguino-scrotal.

Liposarcoma (LPS) is one of the most common soft tissue sarcomas that arises where fat is present. The retroperitoneum and extremities are the most common sites of origin, and up to 40% of liposarcomas are found in the retroperitoneum.[1]

Although relatively uncommon, retroperitoneal liposarcoma (RLPS) can cause substantial morbidity and mortality.[2] Similar to other retroperitoneal soft tissue sarcomas, RLPS is frequently discovered as a giant tumour, often occupying the entire abdominal cavity. Although surgical resection is considered the most effective treatment, RLPS often has a high rate of incomplete resection and local recurrence due to its aggressive growth into vital structures.[3] Detecting signs or symptoms before tumours become enlarged is difficult because the most common symptom is palpating mass, which occurs only when the tumour is large.

Retroperitoneal liposarcoma is usually confined to the retroperitoneum. However, in rare cases, RLPS extends to the inguinal canal, which communicates with the retroperitoneal space. Only a few case reports describe RLPS presenting as an inguinal hernia.[4–9] Sometimes, a protruding mass in the inguinal region is the only symptom, resulting in a misdiagnosis of ordinary inguinal hernia.

Not every inguinal LPS is an RLPS. Similar to RLPS, inguinoscrotal LPS, which arises along the spermatic cord and testis, is an uncommon soft tissue sarcoma.[10] Inguinoscrotal LPS can be isolated in this area without extending into the retroperitoneal space.

The present study summarizes data on LPS of the inguinal region from a high-volume sarcoma centre. We reviewed our experience with inguinal LPS, both RLPS extending into the inguinal region and isolated inguinoscrotal LPS. By comparing these 2 different entities of LPS with the same presenting symptom, we contribute new insights to the understanding of inguinal sarcoma presenting as an inguinal hernia.

Methods

Patients

Data on patients who underwent surgery for LPS located in the inguinal canal between January 1998 and December 2016 at Samsung Medical Center were retrospectively collected from our institution's sarcoma database. Patients were categorized as having RLPS extending into the inguinal region or isolated inguinoscrotal LPS based on tumour location. Only patients with an inguinal LPS on initial presentation were included. Patients with RLPS extending into the thigh region through the femoral canal were also excluded.

Data collection

Demographic data and treatment history from other hospitals were collected by chart review. Anatomic locations of tumours were determined by reviewing preoperative computed tomography (CT) or magnetic resonance imaging (MRI) scans. We assessed laterality, location and size. With respect to surgery, we collected data on resected organs, margin status of the specimen and invasion of adjacent organs. Data on tumour characteristics were histological differentiation and Fédération Nationale des Centres de Lutte Contre le Cancer (FNCLCC) grade, based on the pathology report. Data on adjuvant therapy, such as radiotherapy or chemotherapy, were also collected. The Institutional Review Board of Samsung Medical Center approved our study protocol.

Statistical analysis

We used the Mann–Whitney test to compare continuous variables and the Fisher exact test to compare categorical variables. Kaplan–Meier survival analysis was used to analyze disease-free, overall and inoperable disease-free survival between groups. The date of the operation when curative complete resection was performed was the starting point, and the end point was set as time of interest, recurrence, death, or inoperable progression of the tumour. In cases of inoperable progression, the end point was the time when the team decided that the tumour was inoperable and required only palliative treatment.

We performed all statistical analyses using SPSS software version 18.0 (SPSS Inc.).

Results

During the study period, 179 patients underwent surgery for abdominal LPS at our centre (Fig. 1). Among these patients, 168 (93.9%) had RLPS, 7 patients (3.9%) had inguinal LPS isolated to the area, and 4 patients (2.2%) had LPS in the abdominal wall. Whereas 161 (95.8%) patients with RLPS had a tumour only in the retroperitoneal space, 6 (3.6%) had an RLPS extending into the inguinal canal, and 1 (0.6%) had an RLPS extending into the femoral canal. The 6 patients with RLPS extending into the inguinal canal and the 7 patients with an inguinal LPS isolated to the inguinoscrotal region were included.

Case summary: retroperitoneal liposarcoma extending into the inguinal canal

The 6 patients with RLPS extending into the inguinal canal are described in Table 1. The male:female ratio was 5:1 and the right:left ratio was 1:1. Four patients (66.7%) reported a palpable mass on initial presentation, and 1 had dyspepsia. Patients had a mean tumour size of 27.9 ± 6.8 (range 18–37) cm. The tumour of retroperitoneal origin extended to the scrotum in 2 of 5 patients (40.0% excluding the female patient). Tumour histology was well differentiated in 2 cases (33.3%) and de-differentiated in 4 cases

(66.7%). The CT findings of patients are presented in Figure 2.

Half of these patients (patients 1, 2, and 3) received incorrect diagnoses and had operations for simple inguinal hernia at other hospitals. Curative complete excision of the hidden tumour was delayed for 20, 12 or 62 months, respectively, from the hernia repair. Patient 1 underwent inguinal hernia repair at another hospital. Two cord lipomas, 9 cm and 5 cm, were discovered during the operation. Pathological review was not performed at that time. Patient 2 underwent inguinal mass excision at another hospital. The patient remembered that she was told that the mass was a lipoma. Pathology slides were not reviewed at our hospital. The retroperitoneal mass was discovered after symptoms reappeared 12 months later. Patient 3 underwent hernia repair 5 years before mass excision. Because of a lack of information about the first operation, we could not identify suspicious signs for patient 3. Only patient 2 is currently disease-free, surviving 89 months after excision with no recurrence. Patient 1 and patient 3 died 39 months and 12 months, respectively, after the operation. Patient 3 experienced metastasis to the liver and underwent liver resection.

Three patients underwent mass excision on initial presentation. Patient 5 underwent combined organ resection, including kidney, adrenal and testis resection. The patient did not experience recurrence during the 11-month postoperative period. Patient 4 and patient 6 are currently in an inoperable progression status, even after numerous operations for recurrent tumours.

Case summary: liposarcoma isolated in the inguinoscrotal region

Seven patients with inguinoscrotal LPS are described in Table 2. The male:female ratio was 6:1 and the right:left ratio was 4:3. Five patients (71.4%) sensed a palpable mass and 1 (14.2%) sensed a swelling in the inguinal region. Patient 8 had pain in the scrotal area. Four of 7 patients (57.1%) had tumour involvement in the scrotum. The mean tumour size was 7.8 ± 4.2 cm (range 3–14 cm). Six patients had a first operation in another hospital; 4 came to our centre after recurrence and 2 underwent complete excision in our centre for remnant sarcoma. Two patients (patient 10 and patient 11) underwent combined and mass resection. Five of 7 patients (71.4%) had de-differentiated LPS, 1 (14.2%) had well-differentiated LPS and 1 (14.2%) had myxoid/round cell LPS. Although 5 of 7 patients (71.4%) experienced recurrence, all 7 patients currently have disease-free status. Patient 9 had a lung metastasis that was operated with video-assisted thoracoscopic lobectomy, and was followed up for 173 months. Figure 3 shows initial image findings for patients.

Comparison between RLPS and inguinoscrotal LPS

Comparisons of demographic, clinical and pathological characteristics of patients are shown in Table 3. The RLPS group had significantly larger tumours than the inguinoscrotal group (27.9 ± 6.8 cm v. 7.8 ± 4.2 cm, p =

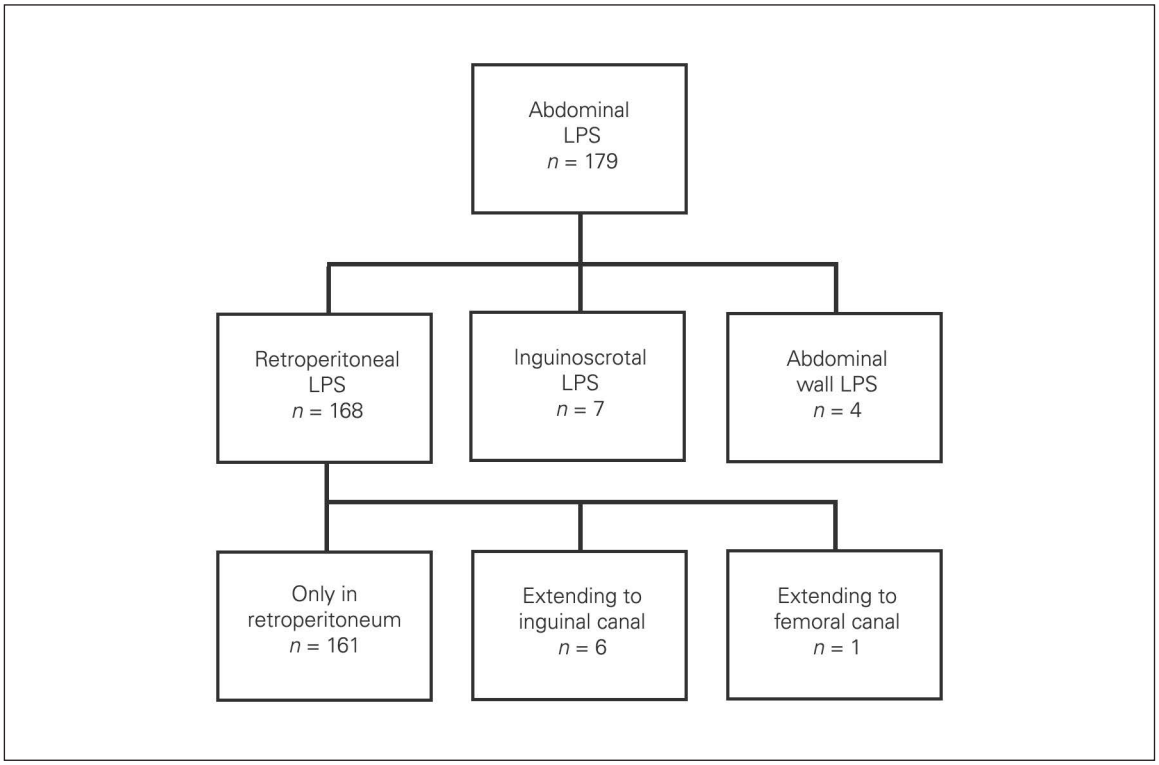

Fig. 1: Patients who underwent surgery for abdominal liposarcoma at Samsung Medical Center. LPS = liposarcoma.

0.001). Postoperative complications were significantly more common in the RLPS group (n = 4, 83.3%). Patients in the inguinoscrotal group experienced no postoperative complications (p = 0.021). The median comprehensive complication index was 8.7 (interquartile range 0–21.72) in the RLPS group, showing a trend that approached statistical significance (p = 0.05). No differences were observed in sex (p > 0.99), mean age (49.7 ± 6.4 yr v. 52.1 ± 12.5 yr, p = 0.37), laterality (p > 0.99), scrotal involvement (40.0% v. 66.7%, p = 0.57), histology (p = 0.52), grades (p = 0.35), positive margin (100% v. 66.7%, p > 0.99), adjuvant therapy (66.7% v. 85.7%, p = 0.56), recurrence (66.7% v. 71.4%, p > 0.99), median recurrence-free duration (12.5 mo v. 18 mo, p = 0.73), or mean survival duration (44.3 ± 36.4 mo v. 71.4 ± 68.2 mo, p = 0.35). Combined organ resection was performed on 83.3% of patients with RLPS, whereas 28.6% of inguinoscrotal patients underwent combined organ resection (p = 0.10). Although 2 patients (33.3%) in the RLPS group died, this was not significantly different from the inguinoscrotal LPS group, in which no patients died (p = 0.19). The proportion of patients who had inoperable tumours was significantly higher in the RLPS group than in the inguinoscrotal group (n = 4, 66.7% v. n = 0, p = 0.021). Inoperable status was defined as death or tumour progression that was considered inoperable.

Disease-free, overall, and inoperable disease-free survival

We performed Kaplan–Meier survival analysis with log-rank tests to compare survival between the 2 groups for recurrence, inoperable progression and death. Disease-free survival in the RLPS group was 66.7% at 1 year and

Table 1. Characteristics of patients who had retroperitoneal liposarcoma extending to the inguinal canal

	Hernia repair as initial operation			Mass excision as initial operation		
Characteristic	Patient 1	Patient 2	Patient 3	Patient 4	Patient 5	Patient 6
Sex	Male	Female	Male	Male	Male	Male
Age, yr	60	52	49	51	43	43
Symptom	Palpable mass	Palpable mass	N/A	Palpable mass	Dyspepsia	Palpable mass
Laterality	Right	Right	Left	Left	Right	Left
Location	Hemiabdomen	Entire abdomen	Pelvis to kidney	Pelvis	Entire abdomen	Hemiabdomen
Tumour size, cm	28	30	18	22.5	32	37
Scrotal involvement	Yes	N/A	No	No	Yes	No
Delay of excision, mo	20	12	62	1	1	1
No. of operations before excision	2	1	1	0	0	0
Excision centre	SMC	SMC	SMC	Outside	SMC	SMC
Resected organs	Mass, kidney, ileuocecum, testis	Mass, ovary, artery	Mass, kidney, colon	Mass	Mass, kidney, adrenal, testis	Mass, kidney, small bowel, colon
Histology	DDLPS	DDLPS	DDLPS 70% WDLPS 30%	WDLPS	WDLPS	DDLPS 90% WDLPS 10%
Grade	3	N/A	N/A	N/A	1	2
Margin	N/A	N/A	Positive	N/A	N/A	Positive
Organ invasion	Positive	N/A	Negative	N/A	Negative	Positive
Postoperative complication (classification)*	Wound (I), delirium (II)	None	None	Ileus (II)	Azotemia(I)	Wound(I)
Adjuvant therapy	Only chemotherapy after fourth operation	Radiotherapy after excision	N/A	Radiotherapy after first excision	N/A	Radiotherapy after excision, chemotherapy after third recurrence
No. of recurrences after excision	2	0	3	8	0	2
Recurrence	Local	N/A	Local, distant (liver)	Local	N/A	Local
Disease-free survival, mo	2	89	1	20	11	14
Death	Yes	No	Yes	No	No	No
Total follow-up, mo	39	89	12	90	11	25
Final status	Death	Disease-free	Death	Inoperable progression	Disease-free	Inoperable progression
Inoperable disease-free survival, mo	28	89	9	90	11	22

DDLPS = de-differentiated liposarcoma; N/A = not applicable; SMC = Samsung Medical Center; WDLPS = well-differentiated liposarcoma.
*Clavien–Dindo classification.

22.2% at 5 years; in the inguinoscrotal group it was 57.1% at 1 year and 42.9% at 5 years. Log-rank tests showed that the groups did not differ significantly ($p = 0.94$).

Overall survival in the RLPS group was 80.0% at 1 year and 53.3% at 5 years; in the inguinoscrotal group it was 100% throughout the study period. Log-rank tests showed that overall survival differences were not significant between the groups ($p = 0.10$).

Inoperable disease-free survival in the RLPS group was 83.3% at 1 year and 41.7% at 5 years; in the inguinoscrotal LPS group it was 100% throughout the study period. Log-rank tests showed significant differences between the groups for inoperable disease-free survival ($p = 0.010$). The 3 survival curves for each group are presented in Figure 4.

Discussion

Retroperitoneal liposarcoma is an uncommon disease that most surgeons do not have much experience managing. Even less common is RLPS extending into the inguinal canal, occurring in 3.6% of patients with the condition, based on our data. In contrast, inguinal hernia is a prevalent condition that all surgeons encounter in clinical practice. The possibility of inguinal hernia being misdiagnosed as LPS is low. Montgomery and Buras[11] reported that the rate of incidental liposarcoma identified during a hernia operation was lower than 0.1% (2 of 1736 inguinal hernias); however, misdiagnosis of uncommon cases can lead to a poor prognosis.

The present study included 6 patients with RLPS extending into the inguinal canal. Previously, only a few published case reports of inguinal hernia were available, and they discussed an LPS protruding from the retroperitoneum.[4–9,12,13] Although 6 is a small number for reliable statistical analysis, we showed that RLPS protruding into the inguinal canal has poor prognosis compared with inguinoscrotal LPS.

One of the important findings of this study was the presenting features of both diseases. First, RLPS extending into the inguinal canal (5 of 6 patients, 83.3%) and inguinoscrotal LPS (6 of 7 patients, 85.7%) were predominant in men. Initial presentation can be similar. Retroperitoneal liposarcoma can remain hidden without signs or symptoms before it becomes large. Extension through the inguinal canal may be the only symptom, and 4 of 6 patients (66.7%) felt a palpable mass in the inguinal region. Three of 6 patients (50.0%) underwent inguinal hernia repair without further checkup (e.g., by CT). One patient had an associated cord lipoma of 9 cm; however, pathology was not reviewed. Another patient underwent associated mass excision and was given a diagnosis of lipoma. Similarly, LPS confined to the inguinoscrotal region presented with a palpable mass or swelling of the inguinal region (6 of 7 patients, 85.7%). Scrotal involvement also occurred in patients with RLPS. Although scrotal involvement was more frequent with inguinoscrotal LPS than RLPS (4 of 6 patients, 66.7% v. 2 of 5 patients, 40.0%), scrotal enlargement with a palpable mass did not

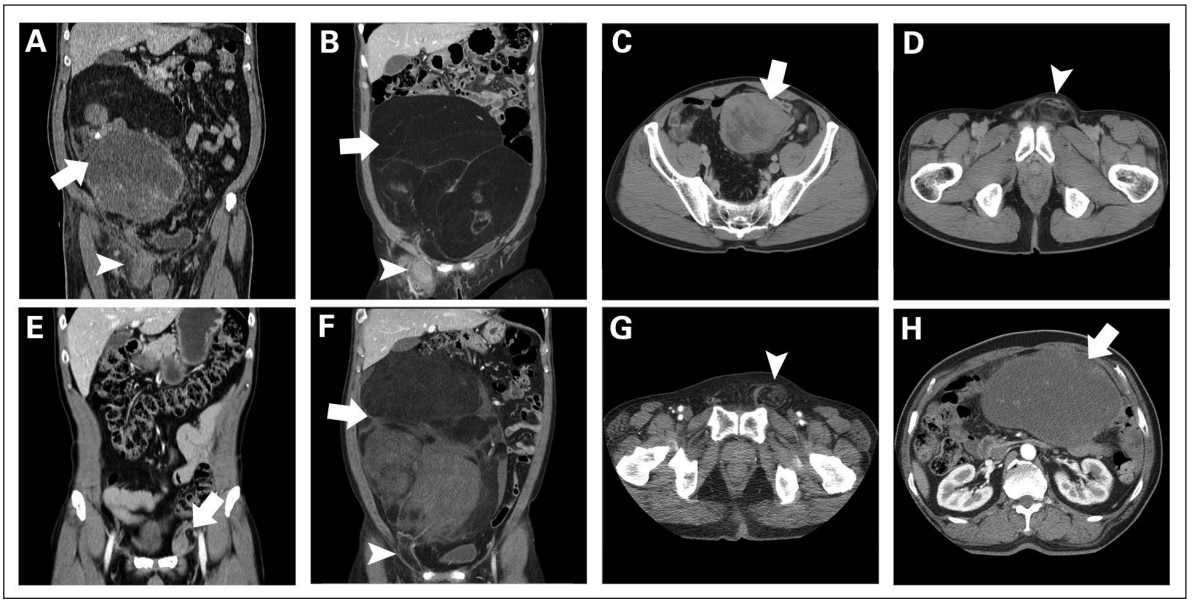

Fig. 2: Computed tomography scans of patients with retroperitoneal liposarcoma extending into the inguinal canal. **A**) Patient 1 had a 28 cm mass occupying the right hemiabdomen with right inguinal protrusion. **B**) Patient 2 had a 30 cm mass occupying the entire abdomen with right inguinal protrusion. Patient 3 had **C**) an 18 cm mass in the pelvis reaching to the lower pole of the left kidney **D**) with extension through the left inguinal canal. **E**) Patient 4 had a 22.5 cm mass in the pelvis with protrusion through the left inguinal canal. **F**) Patient 5 had a 32 cm mass occupying the entire abdomen with protrusion through the right inguinal canal. Patient 6 had **G**) a protrusion through the left inguinal canal **H**) that originated from a 37 cm mass occupying the left hemiabdomen. Arrows indicate the mass of origin, and arrowheads indicate protrusion of the mass in the inguinal canal.

guarantee exclusion of retroperitoneal origin. Based on these presenting features, it was impossible to determine if tumours originated from fat tissue beside the spermatic cord and testis or deep underlying fat tissue of the retroperitoneum. Therefore, imaging studies, such as CT or MRI, can be useful, whereas ultrasonography can be of limited use in scanning the retroperitoneum.

Another finding from this study was the differing prognoses for morbidity and mortality. Based on basic knowledge that RLPS has a poor prognosis, we were not surprised to find that only 2 patients achieved disease-free status (33.3%). Four (66.7%) patients experienced recurrence, 2 (33.3%) patients died, and another 2 patients (33.3%) progressed to inoperable status. The inguinoscrotal LPS group also showed a high recurrence rate (5 of 7 patients, 71.4%). However, patients were all disease-free without progression to inoperable status. Differences yielded by the Kaplan–Meier log-rank test were significant (p = 0.010). The RLPS group showed a high rate of postoperative complications (4 of 6 patients, 66.7%), whereas no patients in the inguinoscrotal

Table 2. Characteristics of patients with inguinoscrotal liposarcoma

Characteristic	Patient 7	Patient 8	Patient 9	Patient 10	Patient 11	Patient 12	Patient 13
Sex	Male	Male	Female	Male	Male	Male	Male
Age, yr	57	59	28	63	61	54	43
Laterality	Left	Right	Left	Right	Right	Left	Right
Scrotal involvement	No	Yes	N/A	Yes	Yes	Yes	No
Symptom	Palpable mass	Scrotal pain	Palpable mass	Swelling	Palpable mass	Palpable mass	Palpable mass, swelling
First operation centre	SMC	Outside	Outside	Outside	Outside	Outside	Outside
Delay of operation, mo	0	0	0	1	0	0	0
Resected organs during first operation	Mass	Mass	Mass	Mass, testis, epididymis, vas deferens	Mass, vas deferens	Mass	Mass
Tumour size, cm	4.5	4	8	12	3	9	14
Histology	DDLPS	DDLPS	Myxoid/round	DDLPS	WDLPS	DDLPS	DDLPS
Grade	2	1	N/A	2	1	1	1
Margin	Positive	N/A	Positive	N/A	N/A	N/A	Negative
Organ invasion	Negative	N/A	N/A	N/A	N/A	N/A	N/A
Postoperative complication	None	None	None	None	None	None	None
No. operations before SMC	0	1	1	1	1	1	1
Reason for operation in SMC	N/A	Completion	Recurrence	Recurrence	Recurrence	Recurrence	Completion
Adjuvant therapy	Radiotherapy after first operation	Radiotherapy	Radiotherapy after second operation, chemotherapy after fourth operation for lung metastasis	Radiotherapy after second operation	None	Radiotherapy after second operation	Radiotherapy after second operation
Recurrence after operation in SMC	Yes	No	Yes	Yes	Yes	Yes	No
Operation after recurrence	Yes	N/A	Yes (VATS lobectomy)	Yes	Yes	Yes	N/A
Total no. recurrences	1	0	6	1	5	1	0
Recurrence	Local	N/A	Local, distant (lung)	Local	Local	Local	N/A
Disease-free survival, mo	7	29	18	8	82	1	47
Death	No	No	No	No	No	No	No
Total follow-up, mo	39	29	173	32	167	13	47
Final status	Disease-free	Disease-free	Disease-free	Disease-free	Disease-free	Disease-free	Disease-free
Inoperable disease-free survival, mo	39	29	173	32	167	13	47

DDLPS = de-differentiated liposarcoma; N/A = not applicable; SMC = Samsung Medical Center; VATS = video-assisted thoracoscopic surgery; WDLPS = well-differentiated liposarcoma.

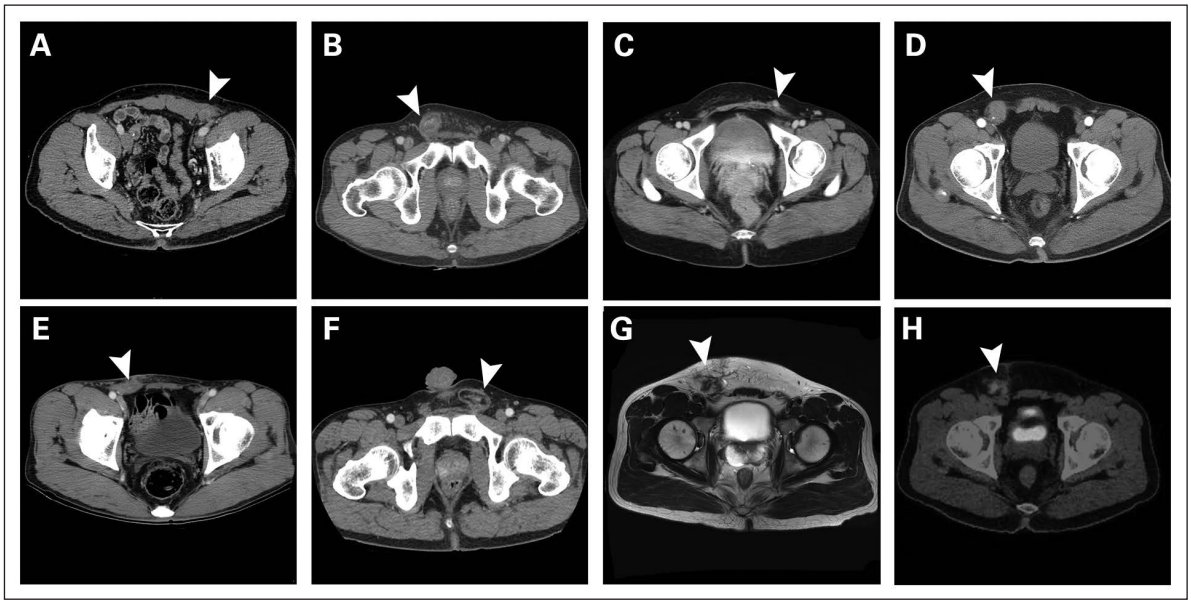

Fig. 3: Image findings of patients with inguinoscrotal liposarcoma. **A**) Patient 7 had a 4.5 cm mass in the inguinal canal. **B**) Patient 8 had a 4 cm mass in the inguinoscrotal region. **C**) Patient 9 had recurrence in the left inguinal canal after previous excision of an 8 cm mass. **D**) Patient 10 had a 12 cm mass in the inguinoscrotal region. **E**) Patient 11 had a 3 cm mass in the right inguinoscrotal region. **F**) Patient 12 had a 9 cm mass in the left inguinoscrotal region. Patient 13 had **G**) a 14 cm mass in the right inguinal canal on magnetic resonance imaging, with **H**) hypermetabolic features on positron emission tomography. Arrowheads indicate a mass in the inguinal canal.

Table 3. Comparison of baseline characteristics and clinical outcomes between patients with retroperitoneal liposarcoma extending to inguinal canal and patients with inguinoscrotal liposarcoma.

	Group; no. (%), or mean ± SD*		
Characteristic	Retroperitoneal LPS (n = 6)	Inguinoscrotal LPS (n = 7)	*p* value
Male sex	5 (83.3)	6 (85.7)	> 0.99
Age, yr	49.7 ± 6.4	52.1 ± 12.5	0.37
Laterality, right:left	3:3	4:3	> 0.99
Scrotal involvement	2 (40.0)	4 (66.7)	0.57
Tumour size, cm	27.9 ± 6.8	7.8 ± 4.2	0.001
Combined organ resection	5 (83.3)	2 (28.6)	0.10
Histology			0.52
Well-differentiated liposarcoma	2 (33.3)	1 (14.3)	
De-differentiated liposarcoma	4 (66.7)	5 (71.4)	
Round/myxoid liposarcoma	0 (0)	1 (14.3)	
Grade (I:II:III)	1:1:1	4:2:0	0.35
Margin positive, %	100	66.7	> 0.99
Postoperative complications	4 (66.7)	0 (0)	0.021
Comprehensive complication index, median [IQR]	8.7 [0–21.72]	—	0.05
Adjuvant therapy	4 (66.7)	6 (85.7)	0.56
Recurrence	4 (66.7)	5 (71.4)	> 0.99
Recurrence-free duration, mo, median (IQR)	12.5 [1.75–37.25]	18.0 [7.0–47.0]	0.73
No. of recurrences, median (IQR)	2.0 [0–4.25]	1.0 [0–5.0]	0.74
Local:distant recurrence	3:1	4:1	> 0.99
Death	2 (33.3)	0 (0)	0.19
Survival, mo	44.3 ± 36.4	71.4 ± 68.2	0.35
Progression to inoperable status	4 (66.7)	0 (0)	0.021
Death	2 (33.3)	0 (0)	0.19
Inoperable progression	2 (33.3)	0 (0)	0.19
Disease-free status	2 (33.3)	7 (100)	0.021

IQR = interquartile range; LPS = liposarcoma; SD = standard deviation.
*Unless indicated otherwise.

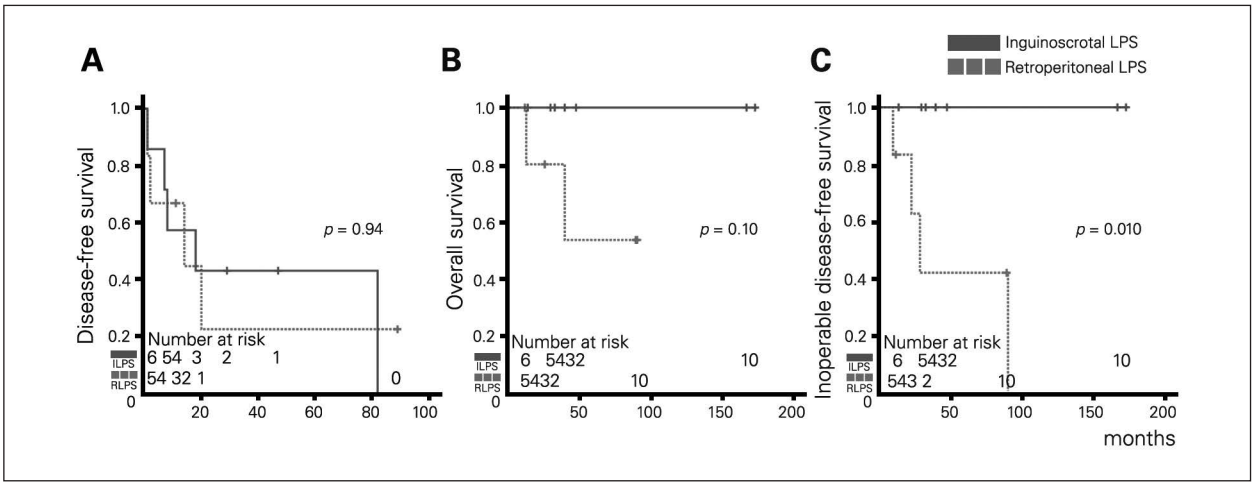

Fig. 4: Survival curves of patients with retroperitoneal liposarcoma (RLPS) extending into the inguinal canal and patients with inguinoscrotal LPS (ILPS). **A**) Groups had no differences in disease-free survival (p = 0.94). **B**) Overall survival did not differ significantly between the groups (p = 0.10). **C**) Inoperable disease-free survival was significantly poorer in the RLPS group than the ILPS group (p = 0.010).

LPS group experienced complications (p = 0.021). Based on these findings, checking for hidden tumours within the retroperitoneal space when inguinal masses are suspected to be soft tissue sarcomas is crucial.

Limitations

A limitation of this study was the small number of patients. However, by discussing a rare circumstance of an uncommon disease, we believe that we can caution general surgeons worldwide who routinely perform inguinal hernia repair. We presented significant findings on tumour size, postoperative complications, inoperable disease progression and inoperable disease-free survival. However, more valuable data could be analyzed if more patients were available for analysis. For example, no significant differences in overall survival (p = 0.10) were observed; these are expected to be different when more patients are included.

Another shortcoming is that we could not perform statistical analysis comparing patients with RLPS who underwent inguinal hernia repair initially and patients with RLPS who underwent complete excision as the first treatment. The importance of not delaying surgical removal of tumours is common sense for surgeons. However, accumulating evidence-based data is still important, even for uncommon conditions.

Conclusion

Retroperitoneal liposarcoma extending into the inguinal canal should be managed with caution. A tumour protruding through the inguinal canal requires high pressure, especially for patients who have already undergone hernia repair. Although no data support this hypothesis, this condition could be associated with worse outcomes, even when RLPS is confined to the retroperitoneum. When a moderate amount of data is collected, comparison of RLPS and RLPS extending into the inguinal canal can be performed.

In our study, half the patients with RLPS extending into the inguinal canal underwent inguinal hernia repair before mass excision. However, the possibility of finding hidden tumours was lost by not performing pathological review in 1 patient's case. Pathological review of every specimen from surgery can provide clues about uncommon diseases. If a mass is large or extends to the retroperitoneal or intraperitoneal space, further imaging workup is mandatory. Protrusion in an already repaired inguinal canal can be a sign of late RLPS. If a mass is detected on imaging, or if pathology shows unusual features, referring the patient to a tertiary sarcoma-specialized centre is important. Retroperitoneal liposarcoma should be resected by an experienced surgeon, and inguinoscrotal liposarcoma can recur if sufficient margins are not achieved during the first operation. We hope our experience and findings provide guidance for general surgeons who frequently operate on patients with inguinal hernias. Furthermore, we hope sarcoma surgeons worldwide present their cases of RLPS extending into the inguinal canal to help researchers generate systematic guidance for this rare condition.

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Affiliations: From the Department of Surgery, Samsung Medical Center, Sungkyunkwan University School of Medicine, Seoul, Korea (Rhu, Cho, Lee, H. Park, J. Park, Kim); and the Department of Pathology, Samsung Medical Center, Sungkyunkwan University School of Medicine, Seoul, Korea (Choi).

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