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W J C C World Journal of Clinical Cases

Contents

Thrice Monthly Volume 10 Number 8 March 16, 2022

OPINION REVIEW

eHealth, telehealth, and telemedicine in the management of the COVID-19 pandemic and beyond: Lessons 2363 learned and future perspectives

Giacalone A, Marin L, Febbi M, Franchi T, Tovani-Palone MR

MINIREVIEWS

Developing natural marine products for treating liver diseases 2369 Wei Q, Guo JS

ORIGINAL ARTICLE

Case Control Study

2382 Analysis of bacterial spectrum, activin A, and CD64 in chronic obstructive pulmonary disease patients complicated with pulmonary infections

Fei ZY, Wang J, Liang J, Zhou X, Guo M

Retrospective Cohort Study

2393 Computed tomography perfusion imaging evaluation of angiogenesis in patients with pancreatic adenocarcinoma

Liu W, Yin B, Liang ZH, Yu Y, Lu N

Retrospective Study

Epidemiological features and dynamic changes in blood biochemical indices for COVID-19 patients in 2404 Hebi

Nie XB, Shi BS, Zhang L, Niu WL, Xue T, Li LQ, Wei XY, Wang YD, Chen WD, Hou RF

Clinical Trials Study

2420 Identification and predictive analysis for participants at ultra-high risk of psychosis: A comparison of three psychometric diagnostic interviews

Wang P, Yan CD, Dong XJ, Geng L, Xu C, Nie Y, Zhang S

2429 Prognostic significance of peritoneal metastasis from colorectal cancer treated with first-line triplet chemotherapy

Bazarbashi S, Alghabban A, Aseafan M, Aljubran AH, Alzahrani A, Elhassan TA

Observational Study

2439 Effect of intraoperative cell rescue on bleeding related indexes after cesarean section Yu YF, Cao YD



Contents

Thrice Monthly Volume 10 Number 8 March 16, 2022

Prospective Study

2447 Effectiveness of the combination of workshops and flipped classroom model to improve tube fixation training for nursing students

Wang YC, Cheng HL, Deng YM, Li BQ, Zhou XZ

META-ANALYSIS

2457 Mortality in patients with COVID-19 requiring extracorporeal membrane oxygenation: A meta-analysis Zhang Y, Wang L, Fang ZX, Chen J, Zheng JL, Yao M, Chen WY

CASE REPORT

- 2468 Escitalopram-induced hepatitis: A case report Wabont G, Ferret L, Houdre N, Lepied A, Bene J, Cousein E
- 2474 Fatal community-acquired bloodstream infection caused by Klebsiella variicola: A case report Long DL, Wang YH, Wang JL, Mu SJ, Chen L, Shi XQ, Li JQ
- 2484 Endoscopic extraction of a submucosal esophageal foreign body piercing into the thoracic aorta: A case report

Chen ZC, Chen GQ, Chen XC, Zheng CY, Cao WD, Deng GH

2491 Severe tinnitus and migraine headache in a 37-year-old woman treated with trastuzumab for breast cancer: A case report

Liu YZ, Jiang H, Zhao YH, Zhang Q, Hao SC, Bao LP, Wu W, Jia ZB, Jiang HC

2497 Metastatic urothelial carcinoma harboring ERBB2/3 mutations dramatically respond to chemotherapy plus anti-PD-1 antibody: A case report

Yan FF, Jiang Q, Ru B, Fei XJ, Ruan J, Zhang XC

- 2504 Retroperitoneal congenital epidermoid cyst misdiagnosed as a solid pseudopapillary tumor of the pancreas: A case report Ma J, Zhang YM, Zhou CP, Zhu L
- 2510 Immunoglobulin G4-related kidney disease involving the renal pelvis and perirenal fat: A case report He JW, Zou QM, Pan J, Wang SS, Xiang ST
- 2516 Fluoroscopic removal of fractured, retained, embedded Z self-expanding metal stent using a guidewire lasso technique: A case report

Bi YH, Ren JZ, Li JD, Han XW

- 2522 Treatment and five-year follow-up of type A insulin resistance syndrome: A case report Chen YH, Chen QQ, Wang CL
- 2529 Effective response to crizotinib of concurrent KIF5B-MET and MET-CDR2-rearranged non-small cell lung cancer: A case report Liu LF, Deng JY, Lizaso A, Lin J, Sun S

World Journal of Clinical Cases		
Contents Thrice Monthly Volume 10 Number 8 March 16, 2		
2537	Idarucizumab reverses dabigatran-induced anticoagulation in treatment of gastric bleeding: A case report	
	Jia Y, Wang SH, Cui NJ, Liu QX, Wang W, Li X, Gu YM, Zhu Y	
2543	Immunoglobulin G4-related disease involving multiple systems: A case report	
	An YQ, Ma N, Liu Y	
2550	Daptomycin and linezolid for severe methicillin-resistant <i>Staphylococcus aureus</i> psoas abscess and bacteremia: A case report and review of the literature	
	Hong XB, Yu ZL, Fu HB, Cai ZH, Chen J	
2559	Isolated scaphoid dislocation: A case report and review of literature	
	Liu SD, Yin BS, Han F, Jiang HJ, Qu W	
2569	Dual biologic therapy with ocrelizumab for multiple sclerosis and vedolizumab for Crohn's disease: A case report and review of literature	
	Au M, Mitrev N, Leong RW, Kariyawasam V	
2577	Cardiac rehabilitation in a heart failure patient after left ventricular assist device insertion and subsequent heart transplantation: A case report	
	Yang TW, Song S, Lee HW, Lee BJ	
2584	Large retroperitoneal atypical spindle cell lipomatous tumor, an extremely rare neoplasm: A case report	
	Bae JM, Jung CY, Yun WS, Choi JH	
2591	Hepatocellular carcinoma effective stereotactic body radiotherapy using Gold Anchor and the Synchrony system: Two case reports and review of literature	
	Masuda S, Tsukiyama T, Minagawa Y, Koizumi K, Kako M, Kinbara T, Haruki U	
2604	Mantle cell lymphoma with endobronchial involvement: A case report	
	Ding YZ, Tang DQ, Zhao XJ	
2610	Fatal systemic emphysematous infection caused by <i>Klebsiella pneumoniae</i> : A case report	
	Zhang JQ, He CC, Yuan B, Liu R, Qi YJ, Wang ZX, He XN, Li YM	
2616	Takotsubo cardiomyopathy misdiagnosed as acute myocardial infarction under the Chest Pain Center model: A case report	
	Meng LP, Zhang P	
2622	Cystic teratoma of the parotid gland: A case report	
	Liu HS, Zhang QY, Duan JF, Li G, Zhang J, Sun PF	
2629	Silver dressing in the management of an infant's urachal anomaly infected with methicillin-resistant <i>Staphylococcus aureus</i> : A case report	
	Shi ZY, Hou SL, Li XW	
2637	Drain-site hernia after laparoscopic rectal resection: A case report and review of literature	
	Su J, Deng C, Yin HM	



Conter	World Journal of Clinical Cases	
Conter	Thrice Monthly Volume 10 Number 8 March 16, 2022	
2644	Synchronized early gastric cancer occurred in a patient with serrated polyposis syndrome: A case report	
	Ning YZ, Liu GY, Rao XL, Ma YC, Rong L	
2650	0 Large cystic-solid pulmonary hamartoma: A case report	
	Guo XW, Jia XD, Ji AD, Zhang DQ, Jia DZ, Zhang Q, Shao Q, Liu Y	
	LETTER TO THE EDITOR	
2657		
2657	COVID-19 pandemic and nurse teaching: Our experience	

Molina Ruiz JC, Guerrero Orriach JL, Bravo Arcas ML, Montilla Sans A, Escano Gonzalez R



Contents

Thrice Monthly Volume 10 Number 8 March 16, 2022

ABOUT COVER

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The primary aim of World Journal of Clinical Cases (WJCC, World J Clin Cases) is to provide scholars and readers from various fields of clinical medicine with a platform to publish high-quality clinical research articles and communicate their research findings online.

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CASE REPORT

Large retroperitoneal atypical spindle cell lipomatous tumor, an extremely rare neoplasm: A case report

Jung-Min Bae, Chang-Yeon Jung, Woo-Sung Yun, Joon Hyuk Choi

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Abstract

BACKGROUND

Atypical spindle cell lipomatous tumor (ASLT) is a rare soft tissue neoplasm with a low potential for malignancy. ASLT frequently occurs in the limb and limb girdles. However, large retroperitoneal ASLTs are extremely rare. There was no concrete case report of retroperitoneal ASLTs.

CASE SUMMARY

An 18-year-old woman presented with abdominal pain and a palpable mass. Abdominal computed tomography revealed a large fatty mass that was approximately 30 cm in size and filled the entire abdominal cavity. Surgical excision was indicated. The tumor did not invade the adjacent organs. The pelvic cavity was then too narrow to dissect smoothly. The mass was successfully excised without tumor rupture or adjacent organ injury. Microscopically, the neoplasm was a well-differentiated adipocytic neoplasm. Immunohistochemical staining showed that the spindle cells were positive for CD34 and desmin, in addition to multifocal positivity for S100 protein. These histological features were consistent with an ASLT. The patient's postoperative course was uneventful. At the 12-mo followup, no evidence of recurrence or metastasis was observed.

CONCLUSION

To the best of our knowledge, our study is the first concrete report of a large retroperitoneal ASLT in the English literature. In the large retroperitoneal ASLT located in the pelvic cavity, which made it too narrow and tight to dissect, complete excision is difficult but very important because of recurrence risk. Although large retroperitoneal ASLTs are considered extremely rare, their



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detection is important for accurate evaluation and management. Owing to their significant rarity, retrospective multicenter case studies are required to determine the clinicopathologic characteristics.

Key Words: Neoplasms; Retroperitoneal space; Spindle cell; Case report

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Core Tip: To the best of our knowledge, our study is the first concrete report of a large retroperitoneal atypical spindle cell lipomatous tumor (ASLT) in the English literature. ASLT is a rare soft tissue neoplasm. Additionally, retroperitoneal ASLT is extremely rare. In the large retroperitoneal ASLT located in the pelvic cavity, which made it too narrow and tight to dissect, complete excision is difficult but very important because of recurrence risk.

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INTRODUCTION

Atypical spindle cell lipomatous tumor (ASLT) is a rare soft tissue neoplasm with a low potential for malignancy or benign characteristics. To date, its etiology remains unknown. ASLT was first described in 1994[1], and for several decades, its diagnosis has been controversial and remains challenging. Recently, a diagnostic consensus has been reached[2].

According to a previous large case study, ASLT frequently occurs in the limb and limb girdles in twothirds of cases, with a predilection for the hands and feet. However, large retroperitoneal ASLTs are extremely rare. Because of the tight pelvic cavity in the present case, surgical excision was very difficult.

In our study, we present the case of an 18-year-old woman diagnosed with a large retroperitoneal ASLT. Despite the surgical difficulty, the postoperative course was uneventful, and no evidence of recurrence or metastasis was observed at the 12-mo follow-up.

We also conducted a review of the literature. To the best of our knowledge, this is the first concrete report of a large retroperitoneal ASLT in the English literature.

This study was approved by the Institutional Review Board (IRB) of Yeungnam University Medical Center (IRB No. 2021-06-020). The patient provided written informed consent for the publication of the case details at admission.

CASE PRESENTATION

Chief complaints

An 18-year-old asian woman presented to our surgery department with abdominal pain, discomfort, and a palpable mass.

History of present illness

About several months previously, this patient felt abdominal discomfort. However, this patient did not further evaluation.

History of past illness

The patient had no other previous medical history.

Personal and family history

The patient had no personal or family history of similar illnesses.

Physical examination

The initial blood pressure was 120/80 mmHg; heart rate, 86 beats/minute; respiratory rate, 14 breaths/minute; and body temperature, 36.9 °C at admission.



Her bowel sounds were normoactive and regular. Physical examination revealed non-specific tenderness throughout the abdomen. However, a very large mass was palpated from the epigastric area to the pelvic area.

Laboratory examinations

The initial laboratory examination revealed normal levels of leukocytes and hemoglobin.

Imaging examinations

Abdominal computed tomography revealed a large fatty mass with a complex soft tissue component. The tumor was approximately 30 cm × 20 cm × 10 cm in size and filled the entire abdominal cavity (Figure 1A). The tumor pushed up the abdominal viscera from the pelvic cavity.

The results led us to consider retroperitoneal neoplasm, including retroperitoneal liposarcoma.

FINAL DIAGNOSIS

Microscopically, the neoplasm was a well-differentiated adipocytic neoplasm with a variable myxoid or collagenous stroma, and the pathologic margin was negative. Delicate and ropey collagen bundles were observed. There was an admixed spindle cell component that showed mild nuclear atypia and hyperchromatia, greater than that usually seen in a benign lipoma, but lesser than that usually seen in a well-differentiated liposarcoma. Immunohistochemical staining showed that the spindle cells were positive for CD34 and desmin, in addition to multifocal positivity for S100 protein, but were negative for MDM2. FISH analysis (fluorescence in situ hybridization) shows no MDM2 amplification (Figure 2). Rb staining revealed positive findings, that is, normal/retained status.

A final diagnosis is ASLT in retroperitoneum based on the immnohistocheminal staining and FISH analysis of the tumor tissue.

TREATMENT

Surgical excision was indicated, and elective exploration was performed thereafter. Laparotomy revealed a huge retroperitoneal tumor. The abdominal organs were normal. The tumor did not invade the adjacent organs. However, it extended to the pelvic cavity and severely attached to the anterior surface of the coccyx and sacrum; the pelvic cavity was then too narrow to dissect smoothly. The peritumoral dissection in pelvic cavity was very difficult and the dissection was performed little by little dissection manner in both lateral pelvic wall surface, anterior coccyx surface and posterior pubic ramus surface. Intra-operative bleeding amount was about 700cc. However, no transfusion was performed in post-operative periods.

Therefore, the surgery was performed for 8 h. The mass was successfully excised without tumor rupture or adjacent organ injury. Gross and microscopic examinations in frozen section biopsy revealed negative surgical margins. The tumor weighed approximately 5000 g, had an egg-shaped appearance, and was 38 cm × 24 cm × 11.5 cm in size (Figure 1B).

The cut surface showed a large fatty mass with diffuse heterogeneous fibrotic change. No hemorrhage or necrosis was observed (Figure 1C).

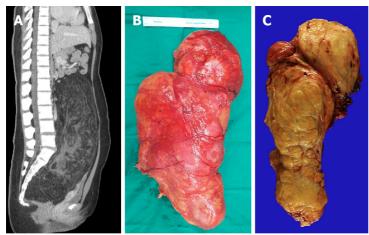
OUTCOME AND FOLLOW-UP

The patient's postoperative course was uneventful, and recovery after surgery was satisfactory; at the 12-mo follow-up, no evidence of recurrence or metastasis was observed (Figure 3). At the time of preparation of this article, the patient is alive and well.

DISCUSSION

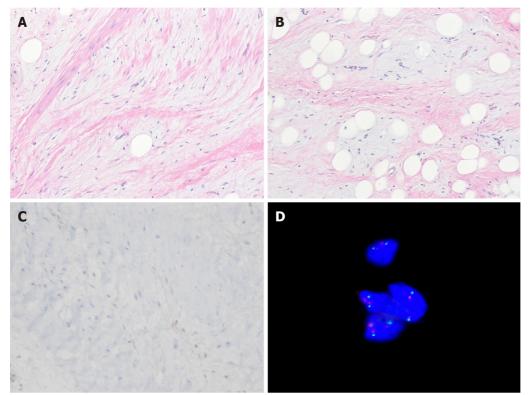
ASLT was first described in 1994[1]. It was previously called spindle cell liposarcoma, differentiated spindle cell liposarcoma, and atypical spindle cell lipoma. Although its diagnosis has been controversial and remains challenging for some time, consensus that ASLT is a subtype of spindle cell lipoma with features different from those of atypical lipoma-like tumor/well-differentiated liposarcoma has been reached^[2]. In 2017, researchers suggested that ASLT and atypical pleomorphic lipomatous tumor (APLT) belong to the same morphologic spectrum, named atypical spindle cell/pleomorphic lipomatous tumor (ASPLT)[3]. Additionally, ASPLT is described in the 5th edition of the World Health Organization (WHO) classification of soft tissue and bone tumors in 2020[4]. Although ASPLT has





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Figure 1 Abdominal computed tomographic findings and gross findings. A: Large fatty mass with a complex soft tissue component. The tumor was filled the entire abdominal cavity and pushed up the abdominal viscera from the pelvic cavity; B: The tumor weigh approximately 5000 g, had an egg-shaped appearance, and mesures 38 cm × 24 cm × 11.5 cm in size; C: The cut surface show a large fatty mass with diffuse heterogeneous fibrotic change. No hemorrhage or necrosis is observed.



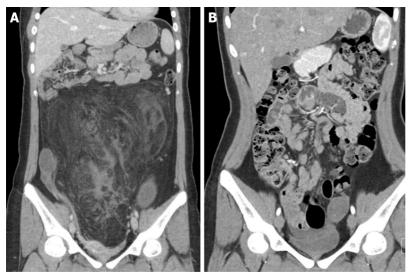
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Figure 2 Microscopic findings. A: The neoplasm shows spindle cells with mild nuclear atypia and hyperchromatia. There are admixed adipocytes (Hematoxylin and eosin stain, × 100); B: The variation of adipocytic size and myxoid stroma are present (Hematoxylin and eosin stain, × 100); C: The spindle cells are negative for MDM2 (Immunohistochemical stain, × 200); D: Fluorescence in situ hybridization shows no MDM2 amplification.

similar clinicopathologic and biological features, ASLT and APLT have different morphological spectra [3].

According to the largest series on ASLTs, the male-to-female sex ratio was 3:2, and the median patient age was 54 years[2]. ASLT develops in the limb and limb girdles in two-thirds of cases, with a predilection for the hands and feet and an approximately equal distribution between superficial and deep sites. They less commonly occur in the head and neck, genitals, and trunk, with very rare retroperitoneal involvement[2].

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Figure 3 Computed tomography. A: Pre-operative computed tomography findings; B: post-operative follow up computed tomography findings. At the 12-mo follow-up, no evidence of recurrence or metastasis was observed.

> The mean size of reported tumors ranges from 5 to 8.5 cm[5]. However, our case developed a tumor in the retroperitoneum with a size of 38 cm.

> Interestingly, the clinical findings of our case, including the patient's age and sex and tumor location and size, are different from the previously published characteristics of ASLT[2].

> The frequent clinical manifestations of ASLT are persistent or enlarged soft tissue masses, nodules, or swelling, sometimes with tenderness. The rare clinical complaints include skin ulceration, local pain, cutaneous vascular markings, cough and hemoptysis, proptosis, night sweats, and abdominal discomfort[2].

> Microscopically, the ASLT in our case consisted of a poorly marginated proliferation of mildly atypical spindle cells set in a fibrous or myxoid stroma, with a variably prominent admixed adipocytic component showing variation in adipocyte size and scattered nuclear atypia, frequently with univacuolated or multivacuolated lipoblasts. Tumor cellularity and the relative proportions of the different components highly varied^[2].

> Although the tumor margins in ASLT are often ill defined with invasion to the surrounding tissues [2], no such invasion was observed in our case.

> The definite diagnosis of atypical adipocytic neoplasm with spindle cell features remains challenging for several decades. Recently, a diagnostic consensus has been reached owing to a better biological understanding with substantial contributions from cytogenetics, molecular genetics, and immunohistochemical correlates^[2].

> CD34 expression is often observed in ASLTs, which is helpful for diagnosis[2,3,5]. Rb protein expression loss is present in 57% of ASLT[2]. In a previous study, the diagnostic sensitivity of CD34 was 64%, and MDM2 expression was not observed in ASLT. Weak and/or focal expression of MDM2 or CDK4 is occasionally present but is always present in the absence of genomic amplification[2].

> Molecular studies have shown deletions or losses of 13q14, including RB1 and its flanking genes RCBTB2, DLEU1, and ITM2B in a significant subset of atypical spindle cell/pleomorphic lipomatous tumors and a consistent absence of MDM2 amplification[2,3,6].

In addition, monosomy 7 has been reported in some cases[7].

In our case, MDM2 expression was negative, and amplification was absent. The negativity of MDM2, CDK4, and FISH for MDM2 amplification highlights critical biological differences between ASLT and dedifferentiated liposarcoma[2,5].

ASLTs can show a wide variety of microscopic features, and differential diagnosis is important and difficult. In our review, differential diagnosis of spindle cell-poor to spindle cell-rich variant of ASPLT was introduced[5]. The spindle cell-associated variants were spindle cell/pleomorphic lipoma, atypical lipomatous tumor/well-differentiated liposarcoma, dedifferentiated liposarcoma, pleomorphic liposarcoma, mammary-type myofibroblastoma, cellular angiofibroma, and solitary fibrous tumors.

The treatment of ASLT involves complete excision, similar to other soft tissue malignant neoplasms. In a large retrospective series, the local recurrence rate was approximately 12%. However, distant metastases were not observed. When complete excision was performed, further oncologic treatment was not required. In previous studies, preoperative or postoperative chemotherapy and radiotherapy were performed in several patients. However, the effectiveness or necessity of chemo-therapy or radiotherapy remains unknown[2].



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ASLT is classified as a benign neoplasm according to the 2020 WHO classification of tumors of soft tissue and bone^[4]. However, several researchers believe that ASLT has a low potential for malignancy or has an intermediate biological potential[2]. Therefore, in our case of a large retroperitoneal ASLT located in the pelvic cavity, which made it too narrow and tight to dissect, complete excision is difficult but very important. Therefore, the surgeon should excise and dissect the tumor carefully without tumor spillage.

According to a large retrospective series, tumor recurrence may develop from 6 mo after the initial surgery; nevertheless, recurrence-related death may not be observed^[2].

As mentioned above, large retroperitoneal ASLTs are extremely rare. In the largest series of ASLT cases, retroperitoneal ASLT was found in only two patients. However, the clinical data of these two retroperitoneal ASLT cases, including the patients' age and sex, tumor size, and prognosis, were not available. Therefore, to the best of our knowledge, our study is the first concrete report of a large retroperitoneal ASLT in the English literature.

CONCLUSION

In conclusion, our study presents a case of a large retroperitoneal ASLT treated with complete excision. In the large retroperitoneal ASLT located in the pelvic cavity, which made it too narrow and tight to dissect, complete excision is difficult but very important. Although large retroperitoneal ASLTs are considered extremely rare, their detection is important for accurate evaluation and management. Owing to their significant rarity, retrospective multicenter case studies are required to determine the clinicopathologic characteristics.

FOOTNOTES

Author contributions: Jung CY, Yun WS, and Bae JM performed the surgery; Jung CY, Yun WS, Choi JH, and Bae JM wrote the manuscript; Choi JH performed the histopathologic diagnosis; Bae JM was the patient's doctor, who revised the manuscript; all authors have read and approved the final manuscript.

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