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Case Report

Rare case of retroperitoneal liposarcoma presented as perineal swelling (The tip of Iceberg)

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Abstract

Background: Primary typical and atypical lipomatous tumors are the most common soft tissue tumors, where liposarcomas are the most common sarcoma. They are mostly found in the retroperitoneum and extremities. Rarely these tumors could present as hernias through different hernial orifices, this case report presents a rare presentation of retroperitoneal liposarcoma presented as a perineal swelling herniating through the ischiorectal and ischioanal fossa.

Case presentation: A 53 years old male patient presented to the outpatient surgery clinic in Al-Hayat National Hospital, Riyadh, kingdom of Saudi Arabia (KSA) complaining of swelling in the left gluteal area for two years, that progressively increased in size with no other symptoms. Magnetic resonance imaging (MRI) revealed a large retroperitoneal soft tissue mass lesion of fat content herniating into the perineal region. Complete surgical excision of this mass were done.

Conclusions: liposarcoma herniating through ischiorectal and ischioanal fossa are rare form of retroperitoneal liposarcoma as in this case where radiology plays an important role in initial diagnosis, staging and surgical planning.

Keywords: retroperitoneal liposarcoma; mri; subcutaneous fat

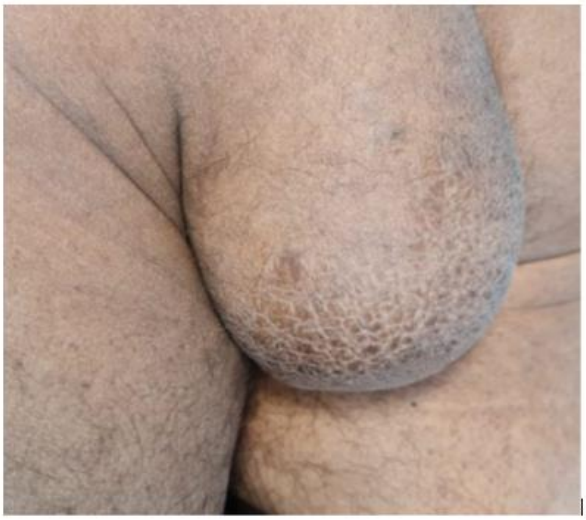
Introduction

Retroperitoneal sarcomas (RPS) are aggressive and rare type of malignant neoplasm, with incidence of about 0.5–1/100,000 inhabitants per year and account for 10–16% of all sarcomas [1]. RPS include heterogeneous and complex group of neoplasia, with four types represent almost 90% of all cases, these are, liposarcomas (LPS), leiomyosarcomas, solitary fibrous tumor, and malignant peripheral nerve sheath tumor [2].

Liposarcomas are the most common type of soft tissue sarcomas, they predominantly occur in the retroperitoneum and thigh [3]. Liposarcomas accounts for 20% of all retroperitoneal tumors and has multiple subtypes, including atypical, well-differentiated and dedifferentiated liposarcomas [4]. Due to the uncontrolled growth in the large potential space of the retroperitoneum, they mostly present with symptoms of mass effect with large median size reaching around 30 cm at the time of presentation making surgical management challenging [5]. Due to the large size of lipomatous tumors, they sometimes present as hernias which can occur through different hernial orifices either in the abdominal wall or in the groin. The herniated part of the tumor represents only the 'tip of the iceberg', as the main part of the tumor is not visualized clinically and is often underestimated, also this late presentation results in higher risk of de-differentiation [6].

Case report

A 53 years-old male presented to the outpatient surgery clinic in Al-Hayat National Hospital, Riyadh, KSA complaining of a swelling in the left gluteal area for two years (figure. 1), that progressively increased in size with no other symptoms. On clinical examination, the swelling was huge in size, soft, not tender not hot with normal overlying skin.



On clinical examination a huge, not tender, pedunculated mass was seen.

Figure 1: photo of the swelling on clinical exam

Ultrasound (US) examination was suspicious for a large gluteal lipoma. The surgeon requested MRI study with contrast for better assessment of the swelling and for pre-operative planning.

MRI was done including axial T1, T2, STIR WIs, coronal T2 and sagittal T2 WIs, then contrast was injection and axial, coronal and sagittal T1 fat suppressed WIs were acquired. On pre contrast images a large well-defined dumbbell shaped mass measuring 11.2 x 9.8 x 31 cm in the AP, TR, and CC dimensions respectively, was seen in the retroperitoneum extending to the left perineal region, filling the lower retroperitoneal/presacral space markedly displacing and compressing the urinary bladder (UB) and prostate anteriorly (fig. 2a), inferiorly it was seen extending downwards at the left ischiorectal fossa, markedly compressing and displacing the rectum to the right side, also this lesion was seen herniating through the left pelvic floor muscles (left levator ani muscle) into the left ischio-anal fossa, displacing the anal canal to the right side and protruding outwards in the perineum (figure 2b). This lesion displayed predominantly hyperintense signal on T1 and T2 WIs that was suppressed in most of the lesion on STIR WIs (figure 3a, b, c). On post contrast images it showed heterogenous enhancing soft tissue components and thick internal septations (figure 4).

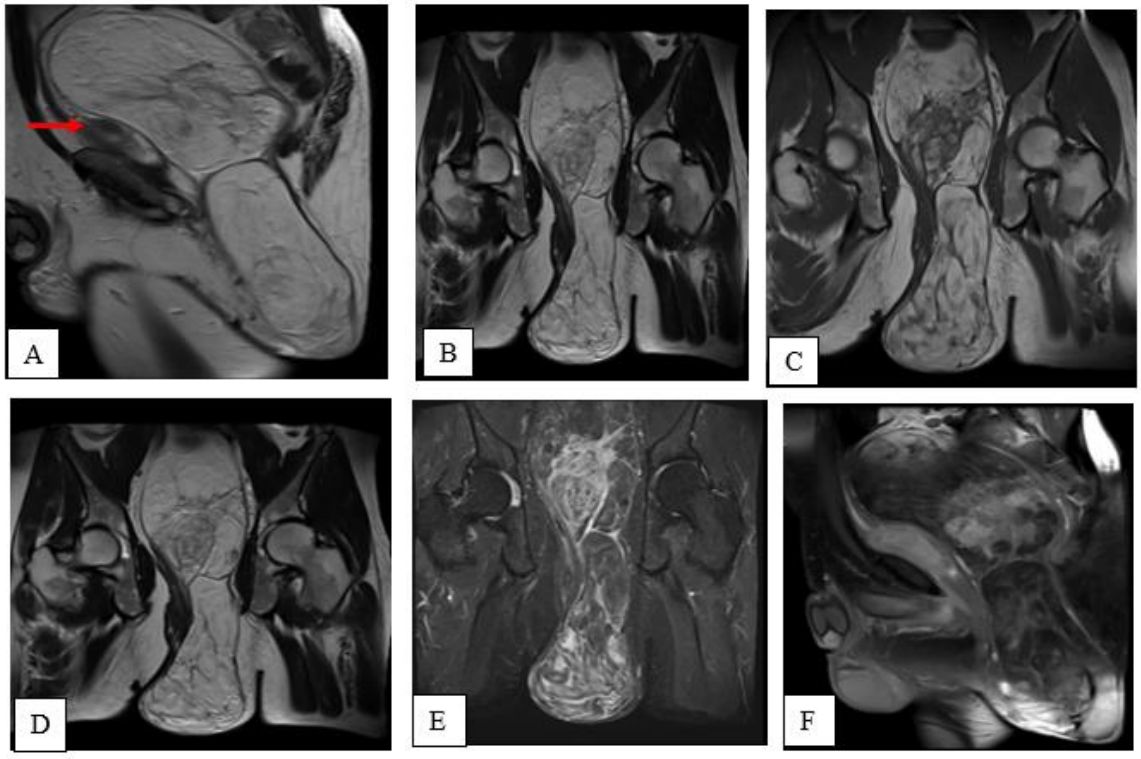


Figure 2: MRI examination of the mass

(A) Sagittal T2WIs shows a large dumbell shaped soft tissue lesion seen filling the presacral space and extending downwards into the perineal region. The prostate is seen markedly displaced and compressed anteriorly (red arrow). (B) Coronal T2 WIs revealed large lesion een extending from the retroperitoneal region inferiorly in the left ischiorectal fossa then through the left pelvic floor muscles (left levator ani muscle) downwards

in the left ischio-anal fossa, to protrude through the perineum.(C) Coronal T1 WIs (D) coronal T2WIs, (E) STIR WIs revealed large soft tissue lesion displaying hyperintense signal in T1 and T2 WIs, most of the lesion is seen suppressed in STIR WIs with internal hyperintense soft tissue components and internal septations. **(F)**Post contrast MRI images of the lesion: Sagittal T1 fat suppressed post contrast images showed internal heterogeneously enhancing soft tissue components and thick enhancing septations.

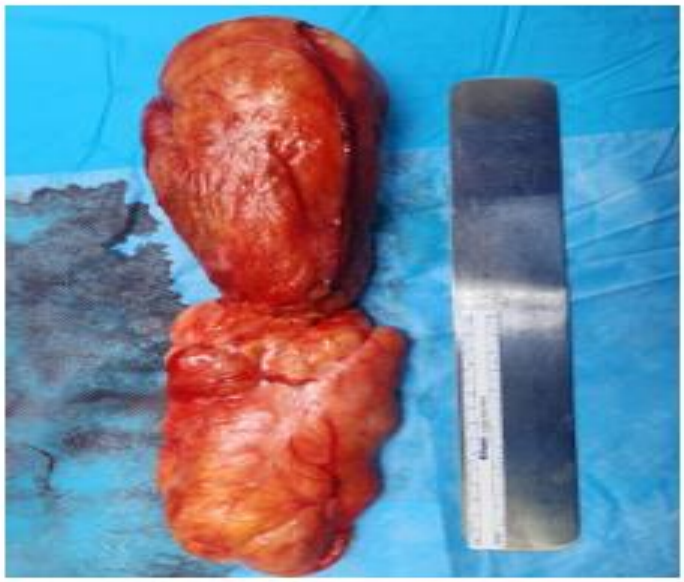


Figure 3: The mass post-surgical excision

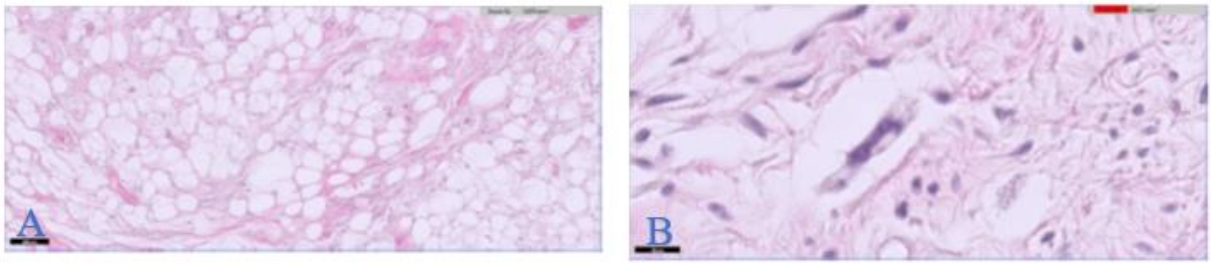


Figure 4: The histopathology of the excised maas show:

A) well-circumscribed hypocellular tumor composed of mature fat plus variably sized adipocytes and fibro-myxoid spindle cells. B) fibrous tissue septae are present containing spindle cells and highly pleomorphic cells, some nuclei have sharply outlined vacuoles (Lochkem) with very few lipoblasts.

The patient was prepared for surgery. On pre-operative assessment, according to The American Society of Anesthesiologists (ASA) classification the patient was ASA1, Exploration laparotomy was through midline incision, the small bowel loops were retracted upwards, the sigmoid colon and rectum were pushed to the right side. Opening of the posterior parietal peritoneum was done and the mass was localized and dissected after securing both ureters laterally. The herniated part was pushed up into the pelvis through retroperitoneal blunt dissection until the mass was completely excised. Hemostasis and then close of the peritoneal defect were done, intra-abdominal suction drain was inserted then the incision was closed in layers (figure 5).

Transversus abdominus Plane (TAP) block was given at the end of operation and before awaking the patient for post-operative pain relief. Reverse given to awake the patient by Sugammadex. No mishaps during the time of anesthesia. After operation the patient was shifted to recovery room for 30 minutes, then admitted to ICU for monitoring of the patient vital signs as he had major surgery and to check his CBC, PH level, ABG and he spent 24 hours there. Histopathology of the excised mass confirmed the diagnosed of well differentiated liposarcoma (fig 6). The patient came for follow up in the surgery clinic which revealed complete disappearance of the large perineal mass. MRI assessment was done one month after surgery which revealed post-operative seroma with no sizable residual tumoral tissue

Discussion

The current case represents one of the rare tumours - giant retroperitoneal well-differentiated liposarcoma that measured 30 cm in the maximal dimension -. Most retroperitoneal liposarcomas are large at presentation; nearly 50% of retroperitoneal sarcomas are greater than 20 cm at diagnosis [7]. Obstructive complications and organ displacement are the most prevalent symptoms that they manifest later [8]. According to the WHO 2020 classification, there are five main subtypes of liposarcomas: atypical lipomatous tumor (ALT)/well-differentiated liposarcoma (WDLPS), dedifferentiated, myxoid, pleomorphic, and myxoid pleomorphic [9]. All subtype of liposarcoma develops in the retroperitoneum. Most of them are dedifferentiated subtypes and well-differentiated liposarcomas. In the retroperitoneum, pleomorphic and myxoid liposarcomas are uncommon. Well-differentiated liposarcoma (ALT/WDLPS) is a mesenchymal tumor that is locally aggressive but does not spread. It is mostly composed of adipocytes and stromal cells, and it has focal nuclear atypia in both of these cell types [10]. Approximately 40–45% of all liposarcomas are ALT/WDLPS. The deep

soft tissue of the trunk and proximal extremities is where it most usually manifests. Moreover, the retroperitoneum is frequently affected as in the present case. WDLPS is a synonym for ALT. Tumor location and resectability are the main factors that determine whether to use either term [10]. Retroperitoneal tumors are extremely difficult to completely excise and are best categorized as WDLPS. Three subtypes of ALT/WDLPS are distinguished histologically: (1) adipocytic (lipoma-like), (2) sclerosing, and (3) inflammatory [11]. The presence of more than one morphological pattern in the same lesion is common, particularly in retroperitoneal tumors. [12]. Dedifferentiated liposarcoma (DDLPS) is an ALT/WDLPS that progresses to a varied histological grade of sarcoma, typically non-lipogenic, either in the primary or recurrent phase [13]. A well-differentiated component may not be found. The high-grade component could sporadically be lipogenic. Roughly 90% of cases arise de novo, while 10% develop in recurrence. The retroperitoneum is the most often affected site of DDLPS. [14, 15 41, 42].

A pleomorphic, high-grade sarcoma with varying numbers of pleomorphic lipoblasts is known as pleomorphic liposarcoma (PLPS) [16 51]. There are no ALT/WDLPS areas or other lines of differentiation seen. PLPS represents a small subset of LPSs, making up less than 5% of all LPSs. [17-19]. Histologically, pleomorphic lipoblasts vary in number within PLPS, which is characterized by high-grade, typically pleomorphic, undifferentiated sarcoma characteristics. For a precise diagnosis of PLPS, lipoblasts must be present [20]. Myxoid liposarcoma (MLPS) is made up of a variety of tiny lipoblasts and homogenous, round to ovoid cells [21 61]. Nearly 20 to 30% of LPSs are MLPSs. MLPSs usually originate in the deep soft tissues of the limbs. About 30–60% of cases develop distant metastases. Primary retroperitoneal MLPSs are extremely rare [22,23 62, 63]. The majority of retroperitoneal MLPS patients are metastatic. According to histology, MLPS is a lobulated tumor with a moderate cell count that is made up of small, ovoid, homogenous cells in a myxoid stroma that has varying amounts of small lipoblasts [21].

When evaluating malignancies that originate in the retroperitoneum, radiologic imaging is essential. It offers helpful data for differential diagnosis (DD) formulation, tumor identification, localization, and characterization, as well as surgery planning [24] Contrast-enhanced computed tomography (CT) is the most accessible and effective imaging technique [25]. Patients with allergies to iodinated contrast agents or with ambiguous involvement in the muscles, bones, or foramina on CT should undergo magnetic resonance imaging (MRI). If radiotherapy (RT) is being considered, MRI may also be helpful for identifying the pelvic disease and evaluating the local tumor extent and surrounding oedema, which are best included when determining the appropriate treatment volume. [26]. Owing to the fluctuations in tumor-grade FDG PET/CT is not routinely employed, it can nevertheless be applied to problem-solving. It is used to assess the possibility of multifocal intra-abdominal disease or when suspicious pulmonary abnormalities are seen on CT [27].

Ultrasound rarely represents a specific diagnosis. Increased echogenecity within the retroperitoneal mass may be due to extensive fatty matrix calcification, increased vascularity, or hemorrhage [28]. On CT, WDLPS is similar in attenuation to macroscopic fat [29]. Liposarcomas on T1-weighted (T1W) and T2-weighted (T2W) MR imaging are isointense to subcutaneous fat with loss of signal intensity (SI) on the fat suppressed imaging sequence [29,7] as was found in the present case, but they differ slightly from normal abdomino-pelvic fat in terms of attenuation, or SI, which aids in the detection of tumor recurrence [30]. WDLPS has inadequate vascularity, which results in little to no contrast enhancement. However, homogenous enhancement may be achieved by small nodular components or thicker septa [29]. Necrosis and calcification areas are rare.

While DDLPS on CT and MRI include areas of nonfatty masses with increased attenuation and low T1W and high T2W SI, they also have areas with attenuation and SI indicative of fat, similar to WDLPS [31]. Ossification or calcification may be observed in 30% of patients [31]. Low attenuation, low signal intensity on T1, and extremely high signal intensity on T2 in relation to muscle are characteristics of the myxoid type on CT and MRI [32]. The myxoid mass may exhibit fat foci. The myxoid subtype's high vascularity causes it to enhance adequately after contrast injection. On CT, pleomorphic and round cell liposarcomas exhibit necrosis, bleeding, and varying enhancement but do not have a fat component. They resemble muscle attenuation [29]. They are heterogeneous on T1W and T2W imaging on MRI and frequently show sites of necrosis and intratumoral bleeding.

DD: Differentiating between peritoneal and retroperitoneal masses can be difficult, especially when large masses alter anatomy. However, displacement of retroperitoneal organs is a useful indicator that a tumor is retroperitoneal in origin [33, 34 31, 32]. Renal angiomyolipoma is taken into consideration if the mass containing fat comes from the kidney and has prominent arteries and renal cortical abnormalities. Due to the bone marrow located inside the fat, adrenal myelolipoma has a frosted glass appearance and is typically better defined than RPLS [35 33]. RPLS should be considered if the mass containing fat is not evidently originating from the solid abdominal viscera. Expansile macroscopic fat external to the solid abdominal viscera is highly suspicious for WDLPS. The presence of septations or solid-enhancing tissue is very suggestive of dedifferentiation. Calcifications may indicate sclerosing or inflammatory variations of WDLPS, or they may suggest dedifferentiation and a poor outcome [36, 37 34, 35]. Benign fat-containing extragonadal dermoids, hibernomas and lipomas may look like RPLS, but they are uncommon in the retroperitoneum. Although fat is not always present in this disease, extra medullar hemopoiesis can also manifest as a mass containing fat [38]. Misdiagnosis most frequently results from a failure to detect abnormal fat [39]. RPLS diagnosis cannot be ruled out just because there is no macroscopic fat present in a retroperitoneal mass. This could be a sclerosing subtype or a disease that has completely dedifferentiated. A diagnosis of a germ cell tumor, melanoma, or metastatic adenocarcinoma may be suggested by a positive serum marker panel or a prior history of malignancy. Measurements of catecholamines in urine may indicate extra-adrenal phaeochromocytoma. Retroperitoneal fibrosis is a rare condition that is taken into consideration, particularly if there is symmetrical ureteric involvement. Lymph nodes are rarely affected by sarcomas, with the exception of epitheliod sarcoma, rhabdomyosarcoma, and clear cell sarcomas. The characteristic imaging picture of retroperitoneal lymphoma is a homogenous mass enclosing rather than effacing vessels. The existence of a sizable, necrotic, retroperitoneal mass that is heterogeneously enhancing and next to a vascular mass strongly suggests the presence of a venous Leiomyosarcoma, which is the second most common sarcoma found in the RP. They are typically derived from the IVC inferior to the hepatic veins; they also originate from smaller vessels such as the renal or, less frequently, the gonadal veins [40]. A sizable, clearly defined, solid vascular tumor, especially one that has obvious feeding vessels, is suggestive of a solitary fibrous tumor (SFT). Lipomatous hemangiopericytoma is a subtype of SFT that contains fat [41]. The retroperitoneum can also harbor benign nerve or nerve sheath tumors. Usually, they are well-defined and rounded, but malignant peripheral nerve sheath tumors (MPNSTs) are an important DD. MPNSTs are frequently the result of neurofibromas, and 50% of cases are associated with type I neurofibromatosis [42]. Certain sarcoma subtypes, including synovial sarcoma, resemble cysts and may be misdiagnosed as hematomas or abscesses due to their characteristic features.

Follow-up imaging: The diagnosis of recurrence can be improved by contrast-enhanced CT surveillance of the chest, abdomen, and pelvis, that can detect recurrences years before symptoms appear. During the first five years, follow-up should occur every three to six months, with yearly imaging after that. Follow-up should continue for at least ten years, if not indefinitely, since the risk of recurrence never plateaus. Routine follow-up's efficacy is not specifically supported by data; however, the strategy is based on general consensus [43]. A CT thorax combined with an MRI

can be used for follow-up, especially for younger patients, who may be concerned about the radiation hazards associated with frequent CT examinations [44]. The gold standard of treatment, particularly for well-differentiated subtypes, is surgical resection. The only effective way to increase patient survival is to completely remove the tumor and any adhering components that was done In the in this case however, it is frequently not achievable because the important structures are frequently affected. Using neoadjuvant radiation therapy post-surgery may help reduce the rate of local tumor recurrence that frequently occurs [28]. Tumor reduction surgery should be carried out even if it is difficult to remove the tumor entirely at an advanced stage in order to lessen compressive symptoms, increase survival time, and enhance quality of life [45 19]. Reoperation is recommended for patients with recurring liposarcomas [46].

Conclusions

Retroperitoneal liposarcomas are rare soft tissue tumors, Familiarity with different imaging findings of liposarcomas and its rare presentations as a hernia through different hernial orifices is critical to ensure accurate preoperative diagnosis as well as proper treatment and surgical planning.

List of abbreviations

(KSA) kingdom of Saudi Arabia, (MRI) Magnetic resonance imaging, (CT) computerized tomography, (US) ultrasound, T1 time 1, T2 time 2, (STIR) short tau inversion recovery, (ASA) American Society of Anesthesiologists, (TAP) Transversus abdominus plane, (ALT/ WDLPS)) atypical lipomatous tumor/well-differentiated liposarcoma, Dedifferentiated liposarcoma (DDLPS), pleomorphic liposarcoma (PLPS), Myxoid liposarcoma (MLPS), malignant peripheral nerve sheath tumors (MPNSTs), solitary fibrous tumor (SFT), Arterial blood gases (ABG), Anteroposterior (AP), Complete blood count (CBC), craniocaudal (CC), Liposarcoma (LPS), Transverse (Tr). Retroperitoneal sarcoma (RPS). Weighted images (WIs).

Declarations:

Ethics approval and consent to participate:

This study was approved by the Research Ethics Committee of the Hayat National Hospitals, the requirement for written consent was waived in this case report and discussed and approved by the ethical committee.

Consent for publication:

Not applicable.

Availability of data and materials:

The datasets used and/or analyzed during the current study are available from the corresponding author on reasonable request**.**

Competing interests:

The authors declare that they have no competing interest.

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Authors contributions:

- Noha M. Taha: Idea of the paper, diagnosis of the case in radiology department, attended the operation to describe exactly the pathway of the lesion and its relations, writing the manuscript.
- Ismail M Ali: writing and revising the manuscript.
- Mohamed I. Ali: The surgeon who make the clinical examination and who did the operation.
- Eldesoky Ibrahim Nouh: The anesthesia doctor who prepared the patient for operation and who made the anesthesia during the operation in the OR.
- Ayed Y. Asiri: The ICU doctor who monitored the patient after this major operation.
- All authors read and approved the final manuscript.

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